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THE PANAMA ADVENTURE

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THE VERY GREAT HONOR of being elected President of this Association carries with it the duty of making an address on this occasion. The selection of a subject, interesting to such a gathering as this, is necessarily a difficult one, and because of this I am venturing something of a departure from tradition, for instead of presenting a technical paper, I shall attempt to tell a simple story of pioneer medical work on the Panama Canal. Much of it may be known to many of you, but it is my hope that its present telling may, nevertheless, be of interest, and appeal especially to those of you who may not have heard it.

Many times I have been asked, or have put the question to myself: "Why do we go into the study and practice of medicine?" Certainly not in the hope of material gain; not for ease and comfort of living; probably not because of a developed missionary spirit. The answer, I believe, should be exactly similar to one given if the question were: "Why does a man go on a Polar Expedition or any other dangerous exploration involving risk and discomfort without hope of any large monetary gain?" To my mind, then, the answer is because the individual has a craving for adventure into unknown realms, a not uncommon type of curiosity. And, after all, can any great geographic discovery or exploration compare with the marvelous explorations and discoveries listed in the history of medicine?

Because I was fortunate enough to have a small part in what I feel was a

great medical project, I shall describe very briefly what might be called "The Panama Canal Adventure."

Since September 25, 1513, when Vasco Nunez Balboa first saw the Pacific Ocean from "a peak in Darien," Panama has been a word which inflamed the curiosity of many men in many lands. The Isthmus has been the pathway toward exploration of the Western Pacific Coast. Across it passed eastward the huge loot of the Incas; Morgan and his pirates struggled through its swamps and jungles to capture and loot old Panama; later it was the path of hope for thousands bound to the gold fields of California; and from the golden days of Philip of Spain men have talked and planned a canal across this narrow strip to unite the oceans.

From its very discovery, this land had been feared as a place of pestilence and sudden death, a fear amply substantiated by every known fact. It has been painted from the earliest writings as a country of swamp and dense jungle filled with crawling and flying death, a country where any white man was sure of swift destruction.

A railroad across the Isthmus was finally undertaken by Americans. It was begun in May, 1850, and its 47 miles completed against unbelievable odds, January 28, 1855. Exact figures of the toll of human life given to this work are unobtainable, but it is a favorite saying in Panama that every cross-tie represents a man lost.

Following his triumphal completion of the Suez Canal, Ferdinand de Lesseps secured a franchise from Colombia, of which Panama was then a part, for the construction of a canal across the Isthmus. In December, 1879, at which time deLesseps was 75 years old, impressive ceremonies were held and the first spadeful of dirt was turned. Owing to various complications actual work was not begun until January 20, 1882. The cost of a completed canal was at that time estimated at $168,000,000, and the time necessary as eight years. The collapse of this great project was evident by the middle of 1886 and was final in January, 1889, after the expenditure of more than $266,000,000 and a conservatively estimated 20,000 lives.

A new French Canal Company was formed, in 1893, to take over the assets and franchise of the old company, and for the following ten years just enough work was done to hold the franchise.

On June 28, 1902, the Spooner Bill was signed by President Theodore Roosevelt. This bill authorized the President to acquire all rights and property of the French company at a cost not to exceed $40,000,000; to secure from Colombia such additional territory and rights as he considered necessary; and to appoint a Commission of seven members to administer the work of construction. On January 22, 1903, the Hay-Herran Convention was signed at Washington, Doctor Herran signing on the authority of the Colombian Government. This treaty stipulated the payment to Colombia of $10,000,000 in gold and in addition the payment of $250,000 yearly in return for the rights and franchises of the French Canal Company and the Panama Railroad, and the ceding to the United States of a strip of land 30 miles in width

extending from ocean to ocean. The Hay-Herran Convention was ratified by the Senate, March 17, 1903. The Colombian Congress met in extra session, June 20, 1903, and finally, after many attempts to secure an enormously increased amount from the United States, the treaty was rejected, August 12, 1903.

On November 4, 1903, the State of Panama revolted against Colombia, and was recognized by the United States as an independent Republic, November 13, 1903. On February 23, 1904, a treaty with Panama on almost exactly the same terms offered Colombia was ratified by Congress. President Theodore Roosevelt appointed the first Canal Commission, February 29, 1904, under the chairmanship of Rear Admiral John G. Walker, and on May 9, 1904, placed the work under the direction of the Secretary of War, then William H. Taft. In his letter to the Secretary on that date, President Roosevelt said: "It is a matter of first importance that the most approved and effective methods and measures known to sanitary science be adopted in order that the health conditions on the Isthmus may be improved. It is the belief of those who have noted the successful results secured by our army in Cuba in the obliteration of yellow fever in that island that it is entirely feasible to banish the diseases that have heretofore caused most mortality on the Isthmus, or at least to improve as greatly the health conditions there as in Cuba and Porto Rico. I desire that every possible effort be made to protect our officers and workmen from the dangers of tropical and other diseases, which in the past have been so prevalent and destructive in Panama."

The logical choice of the man to head this sanitary effort was William Crawford Gorgas, a Colonel in the Army Medical Corps, who had been most successful in the sanitation of Havana, following the announcement of the Commission headed by Colonel Walter Reed of its epoch-making discoveries. Gorgas, as Health Officer of Havana from the fall of 1898, working on the theory that yellow fever was a filth disease, had cleaned up that city as few, if any, cities had ever been cleaned; but in spite of this, yellow fever continued endemically and several sharp epidemics occurred.

The announcement by the Reed Commission, made in February, 1901, completely changed the situation. They presented proof that yellow fever is conveyed from man to man by the bite of the female Stegomyia mosquito (Aedes aegypti). It was discovered that to become infected the mosquito must bite the yellow fever patient during the first three days of the disease; that a period of extrinsic incubation of 12 to 20 days must elapse before this mosquito is able to convey the infection, an important fact worked out some two years earlier by Dr. Henry R. Carter without knowledge of the vector; and that a period of three to six days is necessary before this conveyed infection is manifested by symptoms of the disease in man. An intensive campaign against the Stegomyia was most successful, and Havana, for the first time in 300 years, was free of yellow fever.

This successful work became widely known and the President, the Secretary of War, and the Canal Commission were urged by organized medicine

to put Colonel Gorgas in charge of Panama sanitation. Finally, on June 2, 1904, his appointment as Chief Sanitary Officer was announced and he was ordered to proceed with the work.

Colonel Gorgas was allowed $50,000 for the purchase of supplies, and arrived on the Isthmus, June 28, 1904. On June 30, 1904, the Governor of the Canal Zone, General Davis, issued an order announcing the organization of the Sanitary Department with Colonel Gorgas as its head. This included complete Public Health authority over the cities of Panama and Colon, as well as the Canal Zone. Accompanying Colonel Gorgas, the first party consisted of Medical Director John W. Ross, U. S. N., Director of Hospitals; Dr. Henry R. Carter, U. S. Public Health Service, as Chief Quarantine Officer; Major Louis A. LeGarde, Medical Corps, U. S. A.; Joseph LePrince, Chief Sanitary Inspector; Dr. Ernest Wheeler; Captain Theodore Lyster, Medical Corps, U. S. A.; Surgeon L. W. Spratling, U. S. N.; and Drs. J. C. Perry and C. C. Pierce, U. S. Public Health Service. One week later Dr. Alfred B. Herrick, Dr. Edward P. Beverley, Dr. Arthur I. Kendall and I sailed from New York. Of this original party, only six are now alive. We were followed in succeeding weeks by parties of physicians and nurses, until a working force was built up.

The problem facing the newly created Department of Sanitation was a huge one. The constant sick rate of our army in the Philippines, in 1898, had been above 90 per 1,000, and our Army of Occupation in Cuba, at the same time, reported rates as high as 600 per 1,000. Had it not been for the relatively recent antimalarial work, extending from Laveran to Ronald Ross, and the work of the Reed Commission on yellow fever, the task would have been a hopeless one, for we were to be held responsible for the health of an estimated 50,000 workers stretched across 50 miles of swamp and jungle land, only a short distance above the equator and where the annual rainfall was close to 130 inches, or more than ten feet—literally a mosquito paradise. This 50 miles of jungle was traversed only by the single line antiquated railroad. On the Atlantic side was Colon with about 10,000 inhabitants, and on the Pacific side Panama with a population of about 20,000. Both cities were entirely lacking in any form of sanitation, and even the crudest screening was unknown. The Canal Zone had a population of close to 10,000, living in a number of villages along the line of the railroad, and conditions were similar to those in the larger cities. Early surveys in the Canal Zone showed us that above 70 per cent of the inhabitants were active carriers of malaria and that above 90 per cent harbored the various intestinal parasites from hookworm to ameba. It was immediately apparent that an enormous amount of work and a huge expenditure, as money was then viewed, would be necessary if an adequate working force was to be maintained.

The cities of Panama and Colon and the various villages of the Canal Zone must first of all be furnished with adequate and pure piped water supplies and sewers, if cisterns and water tanks, ideal breeding places for the Stegomyia, the vector of yellow fever, were to be abolished and the intestinal parasitic

diseases overcome. The breeding places of Anopheles, the malaria carrier, over a huge area would have to be abolished by filling and draining swamps and pools, and where this was impossible, by the use of oil and larvicides. Adequate screened quarters would also be necessary for the whole working force situated at proper points to centers of work. Pure food would have to be furnished in a country where practically no food was produced, and ample hospital facilities had to be provided for.

Gorgas faced these enormous problems as perhaps no other man could have done at that time. He may not have been an original investigator, but he had a dogged perseverance that few men possess and he showed excellent judgment in the selection of his aides, notable among them being Dr. Henry R. Carter, a great sanitarian, an authority on tropical diseases, and a man with a true vision that no discouragement could dim. Between these two the major sanitary problems were faced and a program mapped out.

The first Chief Engineer, John F. Wallace, organized a Municipal Engineering Department, which promptly and efficiently began the work of sewer, water supply, and paving construction in the two terminal cities and the main villages of the Canal Zone; but the work of general sanitation was hampered constantly by lack of needed supplies refused or delayed by the first Commission, none of whom apparently believed that such work was necessary and none of whom, except the Governor, lived nearer the Isthmus than Washington. This situation was finally brought to the attention of President Roosevelt by Secretary of War Taft, who had sent a trusted investigator, Dr. Charles Reed of Cincinnati, to the Isthmus, and on March 4, 1905, the first Commission was discharged and a new Commission appointed. Almost immediately the supply situation was remedied, ample materials were forthcoming and many of the troubles of the Department of Sanitation were lessened.

From July through December of 1904, only 13 cases of yellow fever developed. With the advent, however, of many thousands of nonimmunes late in that year and early in 1905, the disease flared up, reaching its maximum in June, with 62 cases for the month, declining to 42 in July and 27 in August, making the total for the year 1905, 233 cases. It is hard to describe the terror created by this disease in the mind of the average individual, but it is sufficient to say that thousands of ablebodied men threw up their work and left on the first available boat. This exodus amounted to more than 90 per cent of the entire American force, and for a time the complete evacuation of all nonimmunes except those in the medical and sanitary forces was considered.

Unstinted praise is due those Americans who, in spite of this panic, stuck to their duties. Such a stand was, of course, expected of the professional group, not all of whom, I regret to say, showed the necessary stamina; but to those members of the engineering and clerical forces who braved exposure and death all credit is due. Among those, I am proud to say, was the young lady who later became my wife, after her recovery from a desperate attack of yellow fever.

Completion of water supplies to the cities of Panama and Colon, together

with unremitting labor on the part of the Sanitary Department, which included the blanket fumigation of the entire city of Panama, accomplished in five days, finally won the battle. Since the single case of yellow fever reported in May, 1906, no further case has occurred on the Isthmus, and except for a slight outbreak of bubonic plague and a sharp one of smallpox, also in 1905, no other terrifying epidemic occurred.

The battle with malaria, however, was a constant one. In spite of unremitting effort this disease remained the major disability problem. In 1906, our malaria peak was reached with a hospital admission rate of 821 per 1,000 employees. This high rate gradually declined until 1913, with a low rate for that year of 70 hospital admissions per 1,000.

Two hospitals had been constructed and maintained by the original French Canal Company: Ancon Hospital was on the outskirts of Panama, built at a cost of close to $3,500,000 and beautifully situated on a terraced mountain side. Colon Hospital was built over a coral reef just out of Colon, overlooking the Caribbean Sea. Ancon Hospital had an original bed capacity of close to 700; Colon about 150. Ancon, finished in 1882, was built in pavilion style, single story, units of 40 beds, of frame, with tile roofs, grouped in units of five and connected by covered walkways, with central operating pavilion and kitchens. It was a show place in the high days of the French, but when the Americans took it over it was sadly run down. From 1889 to 1904, the hospital had been kept up by a devoted group of French Sisters of Charity as best they could. On July 15, 1904, we took it over with Major LeGarde as Superintendent; Dr. Ernest Wheeler as Executive Officer; Dr. A. B. Herrick as Chief Surgeon, but in reality Chief of all professional work; Captain Theodore Lyster, Eye, Ear, Nose and Throat Service; Dr. Arthur I. Kendall, Chief of Laboratories; Dr. Edward Beverley, Assistant Physician; and I, as Assistant Surgeon. Colon Hospital, down to only a few chronic cases, was taken over from the French Sisters in September, 1904, with Surgeon L. W. Spratling, U. S. N., as Superintendent.

We had little equipment at first. The occupants of Ancon Hospital, about 100 chronics, with everything from leprosy to beriberi, were in horrible state. There were no window or door screens. The four posts of each bed were found in shallow water containers as a protection from ants, and each and every one contained larvae and pupae of Stegomyia. Many shrubs in the surrounding grounds were also protected from ants by pottery water containers, all breeding mosquitoes. Colon Hospital was in even worse condition.

Within two weeks American nurses and additional physicians began to arrive and limited supplies were slowly available. Herrick, as Chief of Staff, was wonderful and his rapid grasp of what, to most of us were utterly unknown problems, was marvelous. He had graduated with the second Hopkins class in 1898 and was a finished surgeon and competent physician. He was a tremendous and tireless worker and a hard but wonderfully just taskmaster and teacher. His recent death in Panama deprived me of my greatest friend and benefactor. It is hard in these days to visualize a country in

which no elective surgery had ever been performed, but that was literally true of Panama. Imagine fibroids and cysts of enormous size, old urinary extravasations with dozens of fistulae, lepers of all classes, osteomyelitis cases of years' standing, elephantiasis of every size and variety, herniae with sacs to the knee, and many others, and one has some slight idea of what was poured into the hospital as the American Zone clean-up got under way.

The fever wards were first screened and every patient with temperature elevation was cleared through a receiving ward. In this way early cases of yellow fever were gotten under screening as quickly as possible. Gradually other wards and at last our living quarters were screened.

Within six months of taking the hospital over we were well equipped to handle 600 patients and had built up an efficient staff to handle the constantly increasing number of Canal employees. Within two years Ancon Hospital was enlarged to a patient capacity of about 1,000, including an insane hospital.

The remodeling and rebuilding of Colon Hospital began late in 1904, and was completed early in 1906, with a final patient capacity of 600. Following a short service as Executive Officer to Colonel Gorgas, I was promoted to be Colon's Chief Surgeon in June, 1905. Dr. Groesbeck Walsh was my associate and Dr. Edward Beverley was Medical Chief.

The Canal Zone was divided into 20 medical and sanitary districts, each with an emergency and receiving hospital and dispensary. A well equipped hospital train traveled each way daily, delivering to the two main hospitals all employees needing treatment. No laborer was allowed to remain in quarters if ill and in consequence the hospital admissions were constantly large. We finally had in active use close to 2,000 beds, with a personnel of 102 physicians and 130 nurses, also a very large force of locally well trained Negro orderlies to whom fell a large part of the nursing work.

On June 28, 1905, John F. Wallace resigned as Chief Engineer, to be succeeded by a great man, John F. Stevens. Under Stevens' administration a lock canal, instead of the controversial sea level canal, was decided upon. Railroad problems for the handling of excavated material were solved and laborers were imported in vast numbers—West Indian Negroes, 30,000; Gallegos from Spain, 8,000; Italians, 2,000; and Greeks, 2,000. The vast job was moving, and moving fast, with an *esprit de corps* rarely equaled, and with a well fed, well housed and reasonably healthy force, which, combining men and families, averaged better than 65,000.

Chief Engineer Stevens resigned, April 1, 1907, to be succeeded by Colonel Geo. W. Goethals, U. S. A., as Chairman and Chief Engineer, and a new and final Commission was appointed, of which Colonel Gorgas was a member. Colonel Goethals was the final authority, the other Commission members serving in a purely advisory capacity. He was a great administrator and executive, as well as a great engineer, and under him the large and highly efficient force built up and largely trained by Mr. Stevens functioned smoothly and well.

The "Big Job" was finally finished and opened to world traffic, August 15, 1914, after ten years of work and a total expenditure of $283,500,000 and 6,630 lives. Had the French death rate of 200 per 1,000 per year been maintained, we would have lost at least 78,000 in ten years and would in all probability never have completed the Canal. If we estimate the total sanitation expenditures, including the care of the sick, the injured and the insane, at $20,000,000, have we paid too great a bill for the first great demonstration of what we like to style—Modern Sanitation?

EXTRADURAL DIPLOIC AND INTRADURAL EPIDERMOID TUMORS (CHOLESTEATOMA)

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EPIDERMOIDS (also known as tumeurs perlées, Perlgschwülste, cholesteatome, Epidermoid, and cholesteatomata), arising from the diploe, extradural in position, and those occupying an intradural position are to be considered in this paper. Dermoids, and so-called "cholesteatomata," either primary or secondary, involving the mastoid, arising from the interior of the petrous portion of the temporal bone, accessory nasal sinuses and the bones of the skull, other than the cranium, are not under consideration.

This lesion was rarely found as late as 1920, when Bailey[1] reported two cases of Cruveilhier's tumors (tumeurs perlées) successfully operated upon by Doctor Cushing, in 1919. These two patients were the first recorded cases to recover following removal of the tumor from the intradural position. In 1922, Cushing[2] briefly reported abstracts of six cases of the extradural diploic type appearing in the literature since 1856, with recovery in all six cases. In his paper he reported a case of his own in complete detail. These two papers stand out as the first two milestones of proper surgical eradication of both intra- and extradural lesions.

Since the publication of these two papers, quite a few other cases have been reported from the various neurosurgical clinics of this and other countries. The lesions are still sufficiently rare to permit the author to report six personal cases, and two others from the Neurologic and Neurosurgical Services of Bellevue Hospital, New York.

In 1829, Cruveilhier[3] described three tumors of the intracranial cavity, and on account of their pearl-like appearance gave them the name "tumeurs perlées." One of these cases had previously been reported by Dumeril,[4] and so far as is known, it was the first report of a case of the nonhair-containing type, although one of the dermoid type had been noted by Verratus,[5] in 1745.

In 1838, Johannes Mueller[6] gave the name "cholesteatoma" to this kind of lesion, for the reason that cholesterol crystals were found in his two cases. This apparent misnomer has continued to cling in subsequent papers, and has contributed to the confusion in the minds of many regarding the exact origin and nature of the lesion.

In 1854, von Remak[7] suggested that the tumor arose from embryonic epithelial cell rests, and his work afforded the basis for the conception of the term "epidermoid." In 1897, Bostroem[8] described a definite attachment to the pia mater, and coined the term "haarlosen pialen Epidermoide."

Again, on account of the confusion of the terms used to designate this lesion, Horrax,[9] in 1922, adhered to the general term "cholesteatoma" and

referred to the intradural lesions as meningeal cholesteatoma, stating that: "There can be no mistaking the kind of growth to which reference is made, as this designation immediately eliminates the group of tumors containing cholesterin crystals which arise from pituitary rests, and also does away with any confusion of association with those of the middle ear, cholesteatomata which have not had a primary meningeal attachment."

Critchley and Ferguson,[10] in 1928, suggested that these lesions be termed "cerebrospinal epidermoids." This appellation would hardly include that diploic type which involves primarily the diploe and the *outer* table of the skull.

In 1922, Cushing used the term "epidermal cholesteatoma" to designate the extradural lesion reported. In 1937, Munro and Wegner[11] suggested the term "primary cranial epidermoid" for the diploic type and "primary intracranial epidermoid."

Regardless of the name employed, it is now believed that the tumor, for the most part, is a mass of epithelial débris, which has resulted from slow accumulation of desquamated cells from the epithelial layer of the lining of the lesion. Therefore, it seems that the confusion of terms would be eliminated if the lesion is designated only as an *epidermoid tumor*, either diploic extradural or intradural. In spite of this apparent clear-cut nomenclature, the author has, parenthetically, retained the name "cholesteatoma" in the title of this paper, partly for the reason that almost everyone will more promptly grasp the intent of the subject under discussion, partly due to slowness in giving up the old, and on account of the bibliography.

Incidence.—Epidermoids are comparatively rare tumors, and, therefore, are infrequently found at operation. Previous to the publication of the papers by Cushing and Bailey, the majority of cases were found at autopsy. Since then, more epidermoid tumors have been found and removed at operation, probably because there are more neurologists and neurologic surgeons. More cases have been reported in the last two decades than in all the preceding time. Even so, the percentage of these tumors in neurosurgical clinics is low.

Tooth[12] found but one instance in a series of 258 cases of brain tumor, verified at operation or autopsy, a percentage of 0.4. Frank[13] stated that Bernhardt found only one in a series of 487 autopsies on brain tumor cases, or about 0.2 per cent. In 1920, Bailey reported that only two cases were observed in a series of over 550 verified brain tumors in Doctor Cushing's clinic, a percentage of 0.37. In 1922, Cushing stated that in his series of 740 cases of brain tumor, only three epidermoids were found, less than 0.5 per cent. In 1936, Mahoney[14] reported that in 2,500 verified intracranial tumors in Cushing's collection from Johns Hopkins and Peter Bent Brigham Hospitals, there were 15 epidermoids, 0.6 per cent, and in Foerster's group of 750 tumors, there were five, 0.66 per cent.

It is interesting to note the regularity of the percentage as the number of cases in the Cushing Clinic increased, amounting throughout to about 0.5 per cent.

In 1920, Bailey reported 62 cases from the literature, including two of his own. Mahoney, in his very exhaustive paper published in 1936, reported 142 true cases of epidermoids from the literature, and added five from Foerster's clinic. He did not include the five new cases reported by Bucy,[15] the 11 cases of Love and Kernohan,[16] and small groups, and single cases reported by others.

Location.—As Bailey stated: "The favorite location for these tumors is beneath the pons and midbrain, extending up into the cerebello-pontile angle, but they may occur anywhere on the base of the brain from the anterior perforated space to the foramen magnum." This statement is borne out by Mahoney's statistics, which show that of the 142 verified epidermoids (Table I), only 23 were of the diploic type, and seven were intraspinal. The vast majority, 112 lesions, were intracranial, of which 15 were in the fourth ventricle, 44 were parapituitary and 53 were parapontine.

TABLE I

SERIES OF 142 CASES OF EPIDERMOID TUMORS COLLECTED FROM THE LITERATURE (MAHONEY)

Location	Cases	Sex			Average Age	Clinical Record Available	Psychiatric Symptoms	Operative Results		Autopsy Record
		M.	F.	?				Marked Improvement	Mortality Percentage	
Diploic	23	14	6	3	41 (1–71)	21	3(14%)	17	2(1%)	4
Intra-arachnoid (Intraspinal)	7	5	2	0	25.4(7–35)	6	0(0%)	3	2(40%)	2
Fourth ventricle	15	7	8	0	38.6(19–76)	10	4(40%)	2	4(66%)	9
Parapituitary	44	24	18	2	32.1(5–65)	26	15(60%)	7	8(53%)	29
Parapontine	53	26	22	5	35.3(10–78)	32	20(63%)	2	10(83%)	41
Right	27									
Left	23									
??	3									
Totals	142	76	56	10	35 (1–78)	95	42(44%)	31	26(45%)	85
Intracranial	112	57	48	7	34.5(5–78)	68	39(57%)	11	22(67%)	79

Course.—Wherever located—in the skull, within the cranial cavity, or within the spinal canal—they are slow in growth, due to the very gradual accumulation of epithelial débris within the epithelial lining membrane. However, the slowly increasing pressure exerted by these tumors is irresistible, and all structures give way before it. It is remarkable how a tumor as soft as this can destroy the bone, obstructing its advance, and yet, at the same time, maintain the structural integrity of the soft parts, *i.e.*, the dura, large blood vessels, and nerves.

In three of the cases under consideration the inner table of the skull was completely, and the outer partially, destroyed by large tumors, while the dura and underlying brain were only compressed. In one case, the floor of the anterior fossa was so depressed, and the accessory nasal sinuses so compressed, that the lesion had almost broken through into the nasopharynx, and had perforated the outer table of the skull in the frontal region.

In one instance, the outer table of the skull was absent. The inner table was very thin and depressed, and the pericranium, although elevated, was intact. In another instance, the inner end or tip, and upper half of the petrous

and the homolateral anterior and posterior clinoid processes were destroyed by pressure from a rather large tumor of the middle fossa, while the gasserian ganglion, 5th nerve, and internal carotid artery were still intact. There was also a small cranial defect in the squama, which resulted from compression and erosion. In the single intraspinal case, the soft intradural mass had greatly widened the canal in the lumbar region without destroying the component parts of the cauda equina, and thinning of the dura to any appreciable degree. This was likewise noted by Naffziger.[17] Early recognition and removal of these benign lesions is highly desirable on account of the irresistible and destructive force exerted by them.

Diagnosis.—In the case of the diploic extradural lesion, when the tumor destroys the inner table, it gives the characteristic defect in the skull as shown and described by Cushing, in 1922. In such an instance, the preoperative diagnosis should be made roentgenographically, and complete removal can be accomplished. Cushing stated that so far as he was aware the condition had never been diagnosed except at autopsy or operation. He also stated, however, that "they are capable of recognition roentgenologically owing to the sharp, bony defect, and are capable also of complete surgical removal if approached by a flank, rather than direct attack, in order that the epidermal membrane" may be completely removed.

In 1923, the roentgenograms of the skull of Case 1 (M. D.) were shown to the author without his having seen the patient. The diagnosis was made at once due to the fact that Doctor Cushing's paper had been read shortly after publication in May, 1922, one and one-half years previously. Horrax stated that this was the first time of which he was aware that the preoperative diagnosis had been made. The appearance of the markings about the cranial defect in the two cases was very similar, almost identical, except for size. Therefore, in this instance, one could hardly fail to make the diagnosis.

In Case 2 (J. R.) the defect was small, located in the frontal bone, and involved, for the most part, the outer table, similar to that later shown in Bucy's Case 1. The diagnosis of subperiosteal dermoid was made on account of having seen previous roentgenograms with somewhat similar erosive defects from dermoids. The sharply defined border should have enabled one to make the diagnosis.

In Case 3 (B. M.) the preoperative diagnosis was also made, although the lesion, for the most part, was in the posterior fossa and over the occipital pole, for which reason the cranial defect had similar but not identical markings.

In Case 4 (V. O.) the condition, although suspected, was not diagnosed. The skull markings were not typical, and those present were complicated by remnants of the frontal sinus and ethmoids, which were eroded.

In Case 5 (J. S.), in which an intracranial, intradural lesion was present, one should have made a probable diagnosis instead of that of meningioma, on account of the very slow progress of the lesion as determined by the slow and marked erosion of the petrous tip and clinoid processes, as evidenced in

the groups of roentgenograms made with a nine-year interval, and the very gradual and progressive involvement of the cranial nerves of the middle fossa.

In 1932, Olivecrona[18] ventured a diagnosis of suprasellar cholesteatoma in one case, for the reason that a few days before the patient was seen by him he had operated upon a patient for the same condition, who showed the same ophthalmic and roentgenographic manifestations as the patient in question.

Although in certain instances of intradural intracranial lesions the diagnosis might be made from roentgenographic and neurologic findings, it is believed that in the majority of cases one would be fortunate in making the diagnosis of an operable tumor, and be able to remove it without establishing the preoperative diagnosis, desirable though this may be. Furthermore, the exact diagnosis is not so essential, for it is likely that complete removal of the lesion cannot be accomplished, as in the case of the diploic type.

In Case 6 (D. C.), in which the lesion was in the spinal canal, its location was established roentgenographically in the plain films, later verified by lipiodol, but the nature of the lesion was not diagnosed preoperatively. It is known that other intraspinal lesions can increase the size of the spinal canal. The nature of the lesion was not even suspected; Naffziger's paper had not yet appeared.

In a typical cranial defect produced by the diploic type in which the inner table is more involved than the outer (as observed in Doctor Cushing's notable case and in Case 1), when viewed so that the greatest diameters of the defect are shown, the defect has a *scalloped, dense, clear-cut* margin, showing that this bony margin is more compact than the remainder of the skull. One or more bony hiatuses may be observed in the skull. These represent areas where the outer table of the skull has been completely destroyed. These openings, if they exist, are more apparent on stereoscopic films. Small pieces of detached islands of bone may be seen. The outstanding and differentiating feature of the cranial defect, however, is the sharply defined, dense, white, scalloped margin which is found in no other condition. Any other eroding lesion, regardless of its nature, produces a defect in which the margin is less sharply defined, more hazy, and may be fuzzy and soft.

If the roentgenogram is taken so that one views the defect "on edge" as though looking at a saucer edgewise, a dense line, about 2 Mm. or more wide, will be seen extending from the upper to the lower limits of the defect. This is due to superimposition of the dense margins of the defect which brings the compact bony margins in alignment. The outer table may be so thin that it may not be visible at all in an under- or overexposed film. The characteristics of the defect are positive and unmistakable.

In cases in which the outer table is destroyed, and the inner table still remains or partly remains, the roentgenographic appearance of the cranial defect is not exactly the same pattern as described. Bucy's Case 1 shows the typical defect in this type of case. The bony margin on the lateral view

shows it to be composed of very dense bone, but it is smooth and not scalloped. The marginal, or anteroposterior view, however, shows separation of the two tables of the skull. Some degree of scalloped border may be present, as was shown in Case 2 of this series.

Love and Kernohan, in their report of 11 cases of epidermoid tumor (none of which were of the diploic type) and five cases of dermoid tumors, said that the "extradural and intradiploic epidermoids often produce a characteristic roentgenogram, and, when they do, a diagnosis is not difficult." Going further, they say that "this diagnosis has been made many times by many observers." One can readily agree with the former statement, but the latter is doubtful, unless only a few of the cases observed have been reported. Bucy, in 1935, was able to collect only 13 recorded cases of this type of lesion, two of which were the author's. To this list he added three of his own, which had been operated upon by Sargent, making a total of 16. In 1936, Mahoney collected only 23 cases of this type from the literature.

Appearance of Tumor.—These tumors are definitely encapsulated with a friable lining about 1 Mm. thick. The thickness and color of the membrane vary according to the section removed. In one case (M. D.) the outer surface of the lining membrane was reddish-pink, not unlike the lining of the musculature pulled out of the claw of a boiled lobster. The inner surface, where it came in contact with the caseous mass, was whitish-pink. In the diploic type, where the membrane is in apposition to the thinned-out table of the skull, it can be stripped readily from the bone, or it may come away with the tumor mass. On the other hand, where it is in contact with the dura or pericranium, it is intimately adherent to these structures and cannot be stripped away readily.

In the case of an intradural lesion (J. S.), where the limiting membrane covered the free, protruding portion of the lesion, it was grayish-white in appearance, and no more lustrous than the dura. The outer layer of the membrane did not have the pearly-white appearance, as was seen in the Cruveilhier type of lesion reported by Bailey and others. Its thickness was about equal to that of the overlying dura. Farther down, in the depths, the membrane was intimately adherent to the dura lining the lower, outer and basilar structures and bony confines of the middle fossa, which were much thinner and were almost obliterated.

The beautiful, lustrous, silky, pearly-white appearance of the tumor mass was not observed until its membrane was either broken through or stripped off. The bright mother-of-pearl appearance was more marked in the outer portion of the diploic lesion, where it eroded the bone, than from the inner portions of the tumor, or those which were intradural. Roughly, the outer third of the diploic type mass consisted of pearly-white silky, lamillated material which could be readily split into layers. Bailey accurately described the appearance of this material. He stated: "The surface is smooth, silky, with irregular pea-sized or larger elevations, and peels away easily from the surroundings. The outer surface layers are tough, with about the malleability

of heavy tinfoil." Mahoney described a similar tumor of the diploic type, except that the tumor mass, including the outer layer, was constructed in concentric spirals, like the windings or whorl of a snail. Such an arrangement has not been observed in any of the cases reported here.

The inner portion of the lesion does not present a beautiful appearance. On the contrary, it is composed of an amorphous, grumous, nonhomogeneous mass of crumbling, soft, caseous material, some of which is murky-white, some of which may be a dingy yellowish-brown, and part of it may be brownish-green. Some portions crumble and fall apart when compressed between the thumb and finger, leaving but little substance adhering to the finger and thumb, while other portions are greasy, and stick to the fingers. Although bulky, a given mass of this material is not as heavy as a solid tumor. If an attempt is made to preserve the tumor mass in formalin solution, it disintegrates and falls to pieces and settles to the bottom of the jar as a messy-looking mass of débris. In some instances, a portion of the tumor mass may consist of thick, viscid, greenish or yellowish-green, semisolid, opaque material (Case 4), but this is the exception.

Histologic Structure.—Histologically, the sections of the lining membrane removed and examined in some of the author's cases were almost identical. There may have been a little difference in the relative thickness of the *three layers*, but the sections were so similar that one could believe that all were part of the same specimen.

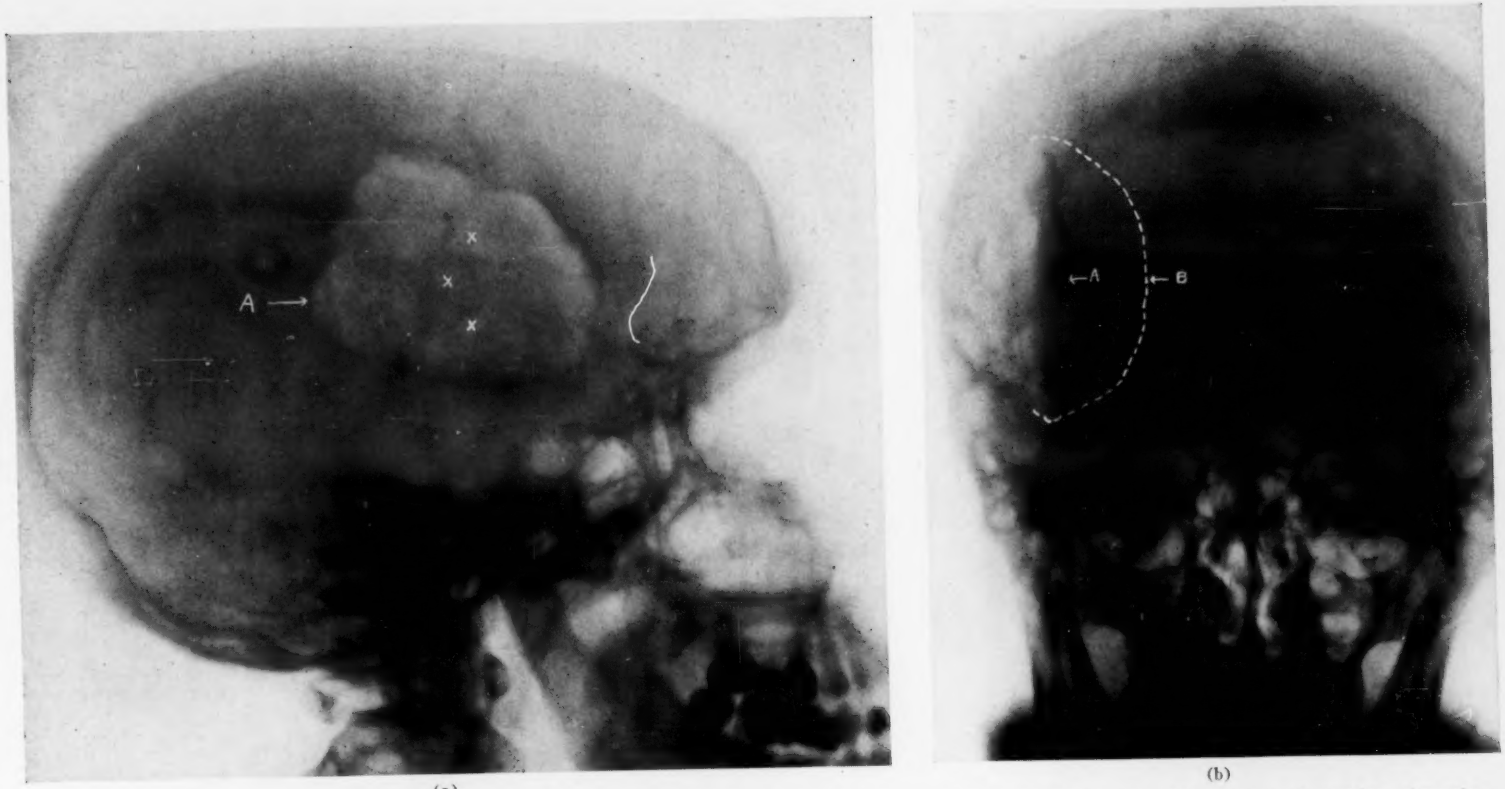
The wall of the lesion is composed of three distinct layers. The outer layer consists of relatively acellular connective tissue which supports the inner layers. It is usually thicker than the second or epithelial layer, but the two may be of the same thickness, depending upon the portion of the wall examined. Within the connective tissue one may see scattered fusiform and stellate fibroblasts in a mass of intercellular fibers. A few blood vessels may be seen.

The second layer, which is internal to the fibrous layer, consists of stratified squamous epithelium. The epithelial layer may be four to 20 cells thick. The cells in the outer portion of the epithelial layer are flattened and are parallel to the surface. These cells have no nuclei. Those composing the inner layer are more cuboidal in shape, and more perpendicular in position. Keratohyaline granules are present in the cytoplasm of these cells. For the most part, the nuclei are well preserved. Intracellular bridges may also be seen.

The third, and most internal, layer consists of cornified epithelium. From this layer inward, the epithelial débris accumulates as the cells are cast off. The lining membrane may be calcified so that the tumor is surrounded by an eggshell-like wall. Such an instance was reported by Horrax,[19] in whose case the tumor removed was enormous.

CASE REPORTS

Case 1.—U. S. Veterans Hospital, No. 81; Reg. No. 1895; *Epidermoid, diploic, extradural, in right temporoparietofrontal region. Preoperative diagnosis. Operation, with complete extirpation. Recovery.*



(a) (b)

FIG. 1(a).—Case 1: Roentgenogram of right cranium showing typical cranial defect with *sharply defined scalloped* border (A). Areas of complete absorption (x). (b) Anterposterior view showing defect with dark line (A), the inner margin of the defect seen edgewise. Approximate inner border of tumor indicated by broken white line (B).

M. D.,* white, male, age 28, single, was admitted November 27, 1923, with the complaints of blurred vision, dizziness and inability to walk well.

Family History.—Essentially negative.

Past History.—Does not know of any illnesses prior to service in army. No accident or operations previous to this time. Had had no spells of nervousness prior to his service.

Present Illness.—While digging trenches with the army in France, during September, 1918, he accidentally fell into a ditch, and on the following day noticed that his right arm and left leg were "paralyzed," and that he had loss of sensation up to the left knee. He had broken his left ankle and was hospitalized continuously until his discharge on certificate of disability, June 1, 1920. He continued to complain of difficulty in using his left leg, and shortly before his admission he noticed that his vision was becoming impaired, and he complained of dizziness.

Physical Examination.—He was well developed, not acutely ill, eyes rather prominent, but no evidence of thyroid disease. Heart, lungs, abdomen and arterial system normal. *Mental Status:* Open, frank, honest, light-hearted, and never worried or brooded much. Previously he always was content with life and made friends easily among both men and women; always got along well with others; sociable, and interested in outdoor sports. Hebrew by faith, but never very religious.

Neurologic Examination.—Pupils reacted well to light, but less to accommodation. All ocular movements, except convergence, well performed. Dalrymple and von Graefe signs positive. Facial movements equal, and tongue protruded in midline. No ataxia. Deep reflexes within normal limits, but somewhat greater on left. Abdominal and cremasteric reflexes equal and active. Walks with a left limp and slight foot-drop. Weakness of flexion of left foot; no clonus or Babinski on either side. Position-sense markedly diminished in left foot and tactile discrimination diminished over left foot. Generalized atrophy from disuse of left lower extremity. Light touch equal on both sides. Possible diminution of pain and temperature sensation over left side of body.

There was a somewhat elevated, visible, and palpable hard swelling in the right temporoparietofrontal region, with slight tenderness on percussion and pressure. There was no pulsation and no doughy feeling. The palpable swelling measured approximately two inches in vertical and two and one-half inches in horizontal direction.

Vision 20/20 both eyes; eyegrounds negative; visual fields normal. Ears, nose and throat negative. Uranalysis, blood count and blood Wassermann negative. Blood pressure normal.

Roentgenologic Examination of Skull.—December 10, 1923: "On *lateral view* (Fig. 1a) there was seen a definite defect in the skull on the right side involving the temporofrontoparietal region. The erosion is clear-cut, and measures in horizontal diameter 7 cm. and in the vertical direction an average of 5½ cm. The superior and anterior borders of the defect are smoother in their definition, while the posterior and inferior margins are more irregular and scalloped. The entire margin, however, is sharply defined and scalloped. A few areas of decreased density are observed, especially near the middle portion of the defect.

"The anteroposterior view (Fig. 1b) also clearly reveals the defect. The outer table of the skull shows irregular erosion in that the table is not of the same thickness throughout, but shows distinct locules with intervening bony projections directed inward, while the external surface of the outer table is smooth, well defined, and free from bony outgrowths or bosses. The erosion of the outer table is greatest near the central portion of the lesion, the table being thicker above and below. There is a distinct linear shadow or line about 1½ or 2 Mm. in thickness, curved slightly inward, running in a vertical direction, with an average distance of 3 cm. from the external surface of the outer table of the skull, and measures approximately 8 cm. in a vertical direction. This is believed

* This case was reported before the Section of Neurology and Psychiatry, New York Academy of Medicine, April 8, 1924.

broken white line (B).

to represent the margins of an eggshell-type of innner table of the skull superimposed on itself.

"Opinion (J. E. J. K.): From the radiographic plates alone, without examination of the patient, it is believed that the lesion present is extradural in origin. It is most likely a cholesteatoma (epidermoid), similar to a case described by Dr. Cushing.[2] It is not believed that the lesion is malignant. The second choice for diagnosis would be an erosive endothelioma (meningioma). This diagnosis, however, is not likely on account of the presence of the definite bony margins about the lesion. Operation for removal is advised."

*Operation.**—January 9 1924: *Crucial incision. Fascia-muscle-pericranial bone flap. "Disclosure of large cholesteatoma." "Separation of tumor from dura and skull." "Complete enucleation." Resection of adherent dura. "Repair of dural defect with fascia lata transplant." Closure.*

The site of the bony defect was outlined on the scalp by actual measurements. The center of the defect was then indicated and a crucial incision was made through the scalp and galea. The four scalp flaps thus marked off—superior, anterior, posterior, and inferior—were dissected up from the temporal fascia and held with two angulated self-retaining retractors. The exposure obtained was fairly circular in shape, with a diameter of about five inches. The retractors gave adequate exposure and prevented bleeding from the edges of the scalp so that it was unnecessary to apply artery clamps or skin clips. The central point of the bony defect having been indicated in the temporal fascia, measurements were made to determine the size of a bone flap sufficiently large to allow of complete removal of the tumor. A horseshoe-shaped incision was made in the temporal fascia, muscle and pericranium, with the base directed downward. A bone flap was made allowing for about a half inch of normal bone beyond the defect. The two most inferior perforations were made well below the inferior border of the defect, and the bone across the base of the flap was cut partly across for a distance of about a half inch from each of the two inferior perforations, so that the flap would break well below the site of the tumor, and not through the bony defect; otherwise there would have been likelihood of tearing through the tumor and its complete removal might not have been accomplished.

The flap, consisting of temporal fascia, muscle, pericranium and bone, was "tilted slightly upward, care being taken in this maneuver that it should fracture well down in the temporal region." "On looking under the slightly raised margin of the flap one could see that the dura was pulled up and was . . . adherent to the internal surface of the tumor." It was possible to separate the dura into two layers, leaving one adherent to the tumor, and a very thin layer adjacent to the cortex. This dissection was tedious. The entire thickness of the dura could have been easily brushed away from the tumor, but on account of the possibility of leaving some of the tumor surface, the dura was split as described above, leaving the outer layer attached to the mass. In making this dissection, the layer of dura adjacent to the cortex was button-holed in three places. Finally, after the dural dissection, or splitting, had been completed, the flap was turned back, carrying with it the tumor, completely dissected away from the dura throughout its extent. "The tumor proved to be of unexpectedly large size and at a late stage of the elevation of the bone flap, the heavy growth began to sag away from its insecure attachment to the bone, so that the whole mass, together with the flap, was held up with a gauze hammock." "The growth was very loosely attached to the irregular, shallow cup it had made for itself in the bone, and was separated without difficulty," except at three small areas. These three areas represented perforations of the outer table of the skull, *i.e.*, the outer table had become completely absorbed at these sites. The largest one was about 1 cm.; the smallest was about 4 Mm. in diameter. All showed on the roentgenograms. At the site of these perforations, the lining membrane of the tumor had to be dissected away and could not be *brushed* away, as was done with the remaining portion of the membrane

* The findings in this case, and the description of the procedure in general were so similar to those in Doctor Cushing's case that verbatim expressions and sentences have been taken from his report and are indicated by quotation marks.

covering the external surface of the tumor. The tumor was not fragmented in its removal, but was completely enucleated. "The outer cup in which it lay consisted of a sharp ridge of bone which projected a few millimeters on to the side of the tumor in its entire circumference, accounting for the distinct peripheral shadow in the roentgenogram." In fact, the tumor, as it rested in the defect in the bone, was somewhat similar in appearance to a prepared overstuffed deviled crab. "This ridge was subsequently rongeured away so as not to leave a projecting edge on the under surface of the skull." "The under surface of the bone showed a very much greater degree of absorption than had been anticipated in view of the roentgenologic findings, there being several irregular areas where no bone whatsoever remained, but merely a dense membrane." The areas on the

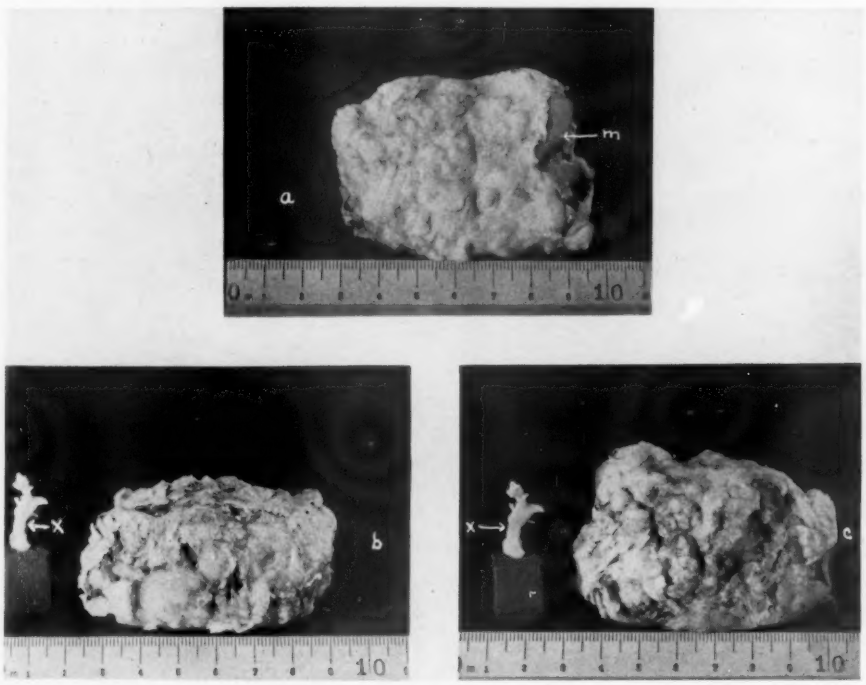


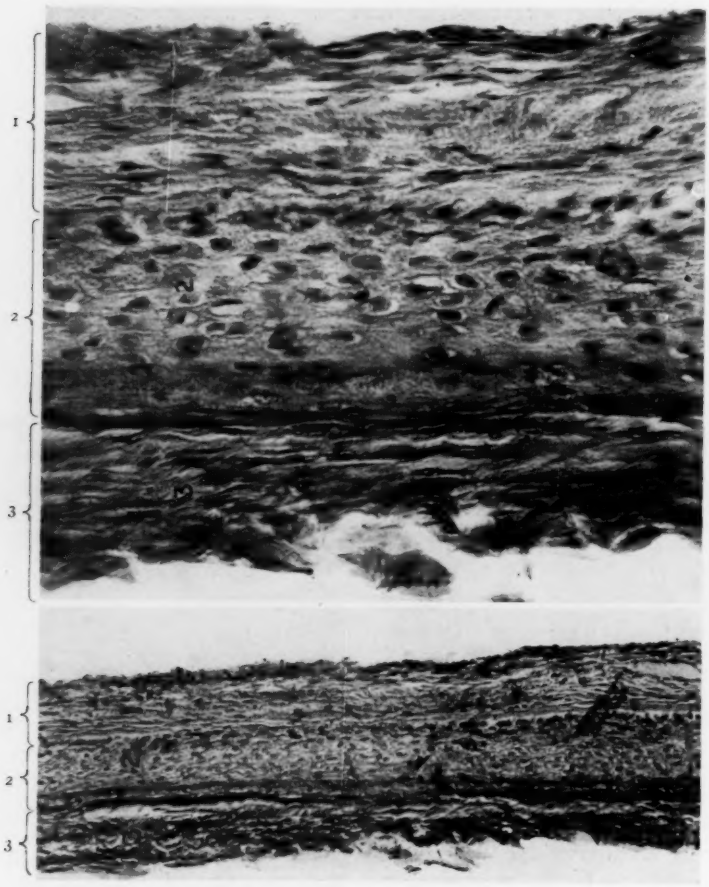
FIG. 2.—Case 1: Photograph of extradural diploic epidermoid. (a) Outer surface. Portion of pinkish-red lining membrane (m) is still attached. (b) Lateral view. (c) Under surface. Piece of skull (x) from scalloped margin of the cranial defect.

under surface of the bone flap, where the tumor was most adherent, were wiped with pure carbolic acid which remained for 15 seconds, followed by alcohol. This precaution was taken to prevent the possibility of recurrence from any area on the bony surface.

The thin layer of dura which remained after the tumor was dissected away fell back and rested in the markedly concave depression in the cortex, but it was not adherent to the cortex. This depressed portion of the cortex did not pulsate, nor tend to become elevated after the tumor was lifted up. On the contrary, it remained depressed like a mashed-in lead pipe. On account of the several perforations in the remaining thin layer of dura, and also on account of the possibility of small pieces of the membrane being left attached, this portion of the dura was resected, leaving a dural defect about two inches in diameter. The defect in the dura was repaired with a fascia lata transplant sutured into the defect with interrupted sutures of chromic catgut No. 00, and a superimposed continuous suture of the same material. Just before the suture was completed, the dura with its fascia lata transplant was floated up from the concave depressed surface of the cortex with saline solution to prevent formation of adhesions between the dura and the

cortex. There was no leakage through the dural suture line. On the contrary, the dura and its transplant remained in a *floated* position. On account of the lack of tendency for the exposed depressed cortex to elevate itself during the operation, it was believed that the transplant might have sufficient time to heal in position in the dural defect before adhesions formed. The bone flap was replaced and the muscle and fascia margins were approximated with interrupted sutures. The scalp flaps were accurately approximated with two layers of interrupted sutures of black silk.

(a)

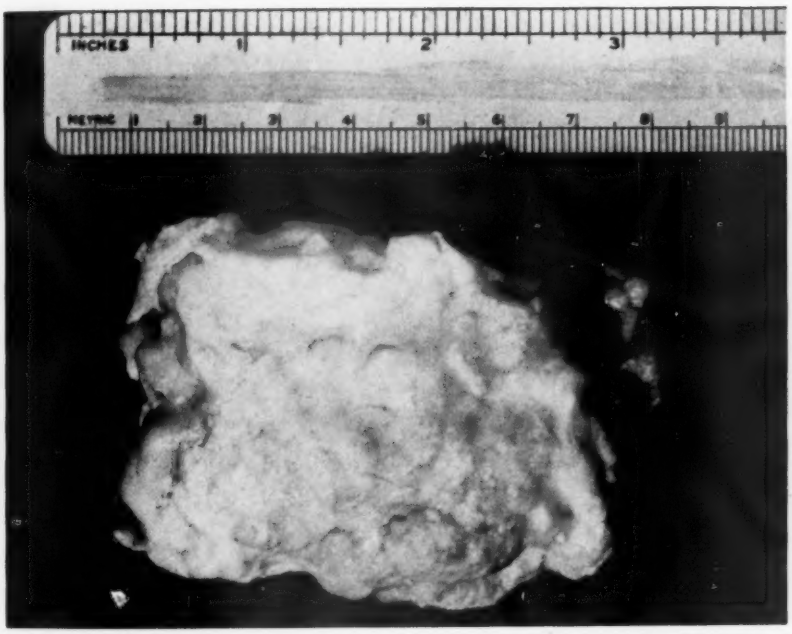


(b)

FIG. 3.—Case 1: Photomicrographs of lining membrane of epidermoid showing the three layers; outer (1), middle epithelial (2), on which the growth of the tumor depends, and inner layer (3). (a) Low power. (b) High power. Keratohyalin granules are present.

This type of incision and exposure was used for the reason that the operation could be more rapidly performed, and also for the reason that the closure was believed

* Photographs and a colored lantern slide of the tumor were sent to Doctor Cushing by the author. In his letter of acknowledgment, March 20, 1924, he stated: "How curious that you should have had one of these cases, and should have diagnosed it after having read my paper! They must be very rare, for I have never seen a similar case, but yours is unquestionably a ringer for it."



Reproduction of a colored lantern slide, made of the gross specimen removed from Case 1. (M. D.)

to be more effective. After this incision is closed, the broad bases of the flaps lie over the margins of the boneflap. Therefore, this closure is more effective than when all of the lines of sutures are superimposed over the margin of the boneflap, as is the case with the typical osteoplastic flap.

Pathologic Examination.—Gross: The tumor (Fig. 2, a, b, c) weighed about 110 Gm., and measured 7x5x4 cm. "Attached to the under surface is a thin fibrous membrane resembling a pseudodura" to which was attached the outer layer of the dura, which was dissected away with the tumor. The outer surface of the tumor and the margins which were in contact with the bone "show the glistening pearly surface of a cholesteatoma" after the membrane was removed. The outer surface, as well as the borders, undulated with elevations and depressions corresponding to the counterelevations and depressions in the bony defect. This picture is most marked on the borders where the roentgenograms showed the typical scalloped effect. The small piece of bone shown in Figure 2 was removed from one of these escalloped borders. The inner surface of the tumor was grayish in color, and was softer than the firm outer portion of the tumor. No hair was present in the tumor mass.

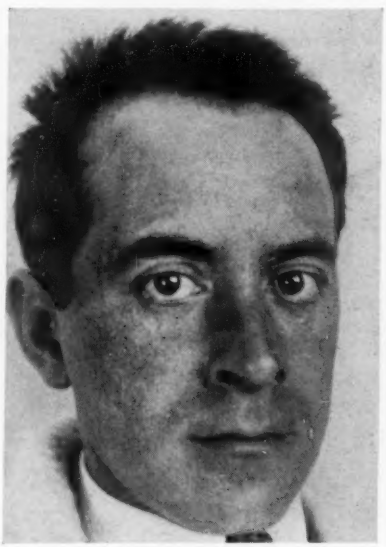


FIG. 4.—Case 1: Photograph of patient two months after operation.

Histologic Reexamination.—Dr. F. Chandler Foot: "The sections show a membrane which has been cut across longitudinally, and is seen to be composed of a heavy layer of connective tissue in which there are some bony spicules and occasional nests of giant cells surrounding acicular spaces of lipid crystals. There are also numerous fat phagocytes in these nests of the 'foam cell' variety. Overlying this there is a layer of stratified epidermal epithelium in which there is a rather poorly defined basal layer, several layers of poorly differentiated cells which probably represent prickle cells and a rather heavy layer of stratum granulosum with deeply pigmented blue granules. Strangely enough, the keratinization of this epidermis is not very marked, although there is a narrow zone of keratinized cells representing the innermost layer of the cyst. Overlying this there are some flakes of desquamated, keratinized epithelium. In some places the keratinization is more marked than in others. The wall of this cyst is totally devoid of any of the adnexa of the skin; there are no hairs, sweat or sebaceous glands present, therefore, the cyst is of the epidermal variety. *Pathologic Diagnosis:* Epidermoid (cholesteatoma)."

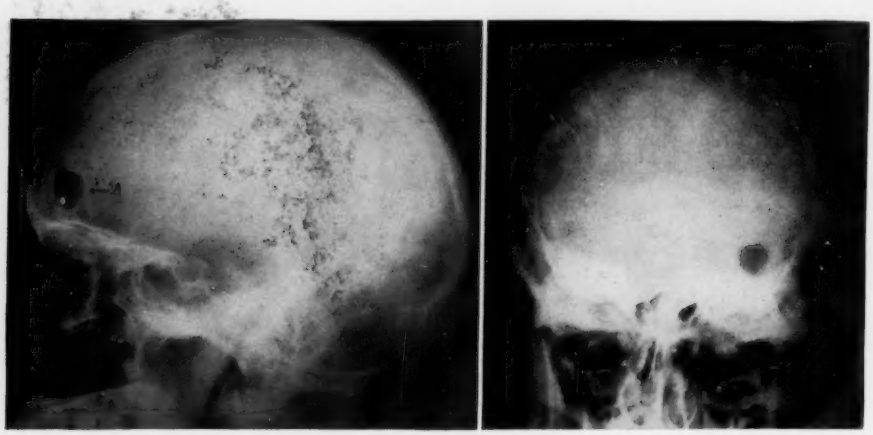
Postoperative Course.—Recovery was uneventful. The wound healed without infection, and without cerebrospinal leakage. On January 20, 11 days after operation, the patient was up in a wheel chair and stated that he was feeling fit. The apparent exophthalmos had become much less. He had no complaints. The dizziness disappeared, and speech was faster than before operation. The slight slowness in thinking had disappeared. He has remained well since, so far as could be learned. Photograph made two months after operation.

COMMENT.—This case has been reported in detail for the reason that it so completely parallels Doctor Cushing's case. The diagnosis was made from his description of the roentgenograms and the lesion. One should encounter but little difficulty in making the diagnosis in a similar case. Had roentgeno-

grams of the skull been made in 1918, when some were made of the foot, the cranial defect would have been found, and the tumor could have been removed five or six years earlier.

It is surprising how such an enormous mass could be present with so few neurologic findings. It must be due to the fact that the lesion, which obviously increases very slowly in size due to its inherent nature, allows the brain to accommodate itself to the encroachment of the tumor. This is in marked contradistinction to the rapid displacement of the brain from an extradural hemorrhage.

Case 2.—*Epidermoid (cholesteatoma); diploic; small. Destruction of outer and depression of inner table of skull. Palpable mass. No neurologic findings. Complete extirpation. Recovery.*



(a) (b)

FIG. 5.—Case 2: Roentgenograms showing small cranial defect produced by the smallest epidermoid of series. (a) Lateral view shows definite clear-cut border posteriorly (A) where the rays strike perpendicularly. Anterior margin not so definite. (b) Anteroposterior view. Postero-anterior exposure would have shown a more sharply defined margin. Outer table was destroyed, inner table depressed.

J. R., male, age 27, was admitted to U. S. Veterans Hospital No. 81, in February, 1925. There was a small doughy mass about the size of a hickory nut in the left frontal region, one inch above the supra-orbital ridge. No signs or symptoms other than the presence of a small mass which resembled a firm, fixed sebaceous cyst.

Roentgenologic Examination (No. 6882).—Figure 5, a and b, revealed the presence of of a small cranial defect in the left frontal region about 2 cm. in diameter, fairly circular in shape except for a notch on the antero-inferior border of the defect. The margin of the defect presented a narrow, dense line posteriorly, similar to that seen in the typical defect made by an intradiploic epidermoid, with scalloping at only one place. The outer table was completely destroyed, while the inner table was intact but slightly depressed. In retrospect, one should have made the diagnosis before operation. The lesion was so small and its position so near that of the common site of a paracanthus subpericranial dermoid that the diagnosis of a dermoid was made.

Operation.—Under local anesthesia, the lesion was completely shelled out of its position in the bony defect. The outer table of the skull was absent, as in Bucy's Case 1, while the inner table was thinned-out and depressed. It was not perforated. The mass measured about 2 cm. in diameter, was encapsulated and was fairly circular in

shape. It was doughy to touch and was set in the cranial defect like a bullet, flattened on the outer side, in half of a bullet mold.

Pathologic Examination.—Gross: Dr. D. S. J. Jessup. "There is a thin-walled sac two centimeters in diameter, with its contents made up of white flaky masses which are very soft and crumble. Section taken through the cyst wall is of paper-like thinness. *Microscopic.*—Section through this limiting membrane shows it to be lined by cuboidal or flattened epithelium attached to which are desquamating cells. The detached material in the tumor appears to be made up of the same kind of cells rolled up on themselves. The white flaky soft tissue débris shows many flat particles of cholesterol mixed with colorless, round bodies which appear to be desquamated epithelial cells. No hairs or glands were noted. *Pathologic Diagnosis:* Cholesteatoma (epidermoid) of frontal bone."

The clinical record could not be obtained from the hospital, so that hospital number and other data cannot be given. Only a few notes, the roentgenograms, and pathologic report were in the possession of the author.

COMMENT.—So far as is known, this is the smallest epidermoid to be reported. The diagnosis should have been made from the slight doughy feeling and from the examination of the roentgenograms.

Case 3.—Bellevue Hospital, Acc. No. 34971-79: *Epidermoid (cholesteatoma); extradural; diploic; occipital; left. Complete removal of caseous mass by morsellation and curettage. Incomplete removal of lining membrane. No infection. Recovery.*

B. M., white, male, age 26, was admitted to Neurologic and Neurosurgical Service, April 21, 1934, with complaint of constant headache for one year.

Family History.—Irrelevant.

Past History.—Negative, except that the patient was told by his mother that, at age seven, he suffered from "some disease" which made it impossible for him to move his neck well for about six months.

Present Illness.—The patient complained of constant headache for the past year. These pains radiated to the right occiput and toward the left eye, and became generalized. Stooping intensified the pain. For about a year he had noticed a swelling about the size of a "small walnut" in the left occipital region. No history of head trauma.

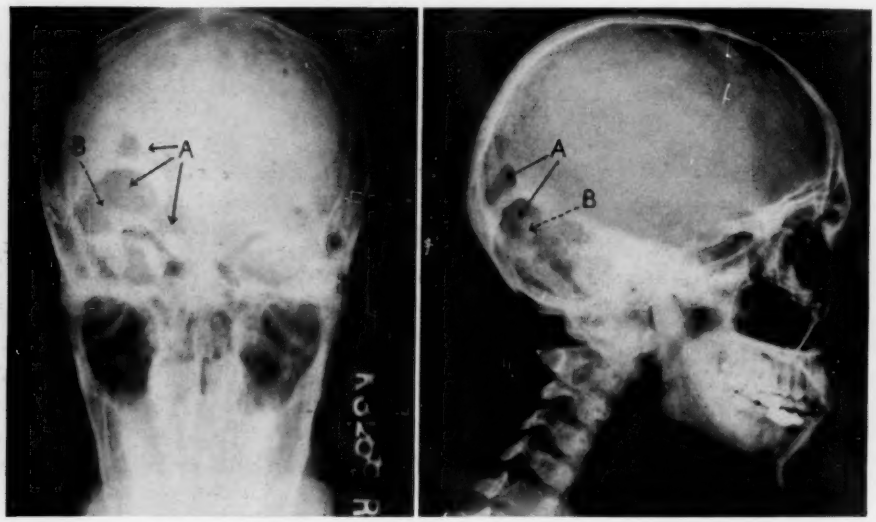
Physical Examination revealed a well developed young man, not acutely ill. General examination negative. In the left occipital region there was an irregular, firm, walnut-sized elevation, about 2½ cm. in diameter, regular in outline and apparently fixed to the skull. It was bony hard, except in its central portion, which was slightly doughy. The scalp was freely movable over it, and it was not tender. T. P. R. normal. Blood count and blood pressure normal. N.P.N. 37; blood sugar 60, blood cholesterol 110. Lumbar puncture not advised.

Neurologic Examination.—Left pupil slightly larger than right. Both reacted well to light and accommodation. Fundi showed definite blurring of both disk margins. Visual fields grossly normal. External ocular muscle normal. Other cranial nerves normal. Motor coordination, reflexes and sensory status normal.

Roentgenologic Examination (Fig. 6, a and b).—There was a cranial defect in the occipital bone on the left side which extended from just behind the posterior margin of the mastoid process, without involving it, to the midline, and measured 7½x8 cm. Near the upper limit of the defect there was a narrow bridge of bone of the outer table separating the upper from the lower portion, so that the upper portion was an elongated oval defect separated from the major defect. The bony margins were scalloped, dense and firm, similar to those seen in films of typical cases of extradural diploic epidermoids. This diagnosis was made.

Operation.—May 1, 1934: Avertin and local anesthesia. A straight, vertical incision was made about halfway between the external occipital protuberance and the

mastoid process, directly over the central portion of the palpable tumor, and extending above and below it. Counter incisions were made about the midportion to afford better exposure. The scalp flaps were dissected up from the fascia and held with self-retaining retractors. Good exposure, practically no bleeding. A horseshoe-shaped incision, with the base directed downward, was made in the fascia and musculature overlying the lesion. When the pericranium was stripped away, perforations in the bone were seen. A bone flap could not be turned. The defect in the skull was enlarged until it measured about two by three inches. On palpation, the tumor offered resistance like that of fairly firm rubber or stiff dough. The mass was definitely encapsulated by a membrane of grayish color. When a portion of this thin membrane was stripped away, the surface of the underlying mass presented the typical pearly-whiteness of the outer layer of an epidermoid.



(a) (b)

FIG. 6(a).—Case 3: Anteroposterior and (b) lateral roentgenograms. Areas of complete absence of outer table (A) with bridge of bone between. Island of bone (B).

Mechanically, it was impossible to remove the tumor in one mass, as was accomplished in Cases 1 and 2; therefore, it was morsellated and scooped out with a brain spoon. The innermost portion of the mass was cheesy in consistency and was of a dirty grayish-green color. The lesion extended from the lateral sinus on the outer side to the midline and from about three-quarters of an inch superior to the lateral sinus to the posterior margin of the foramen magnum (Fig. 7, a). The exact thickness of the tumor could not be determined at operation, but on the lateral roentgenologic view it appeared to be about two inches thick. During removal of the caseous material, as successive scoopfuls of the tumor mass were removed, the floor of the excavation rose toward the skull. After complete removal of the contents of the lining membrane, the dura covering the left cerebellar lobe bulged outward so that it completely filled the space previously occupied by the tumor. This was quite opposite to the observation made in Case 1 in which the dura remained depressed. It was believed that the tendency to extrude the caseous mass through the defect and the elevation of the dura into the defect after removal of the tumor were due to release of compression on the aqueduct which allowed filling of the subarachnoidal space in the posterior fossa. The lateral sinus was seen throughout its course from the mastoid process almost to the torcula. It was about three-sixteenths of an inch wide, markedly compressed, and covered with

a thin, whitish membrane which lined the dura. Complete removal of the membrane was not attempted for fear of injury to the sinus.

Bony islands from the outer table of the skull, adherent to the undersurface of the pericranium, were removed and the flaps were returned to position and sutured in layers. The undersurface of the pericranium came into apposition with the cerebellar dura which previously had been depressed about two inches. The morsellated fragments of the tumor weighed 110 Gm. Throughout the entire extent of the lesion, on the external surface, there was a layer of white, pearly, lamillated material about three-sixteenths of an inch thick, while the inner surface of the lesion was not covered with this white, pearly material.

Pathologic Examination.—Gross: Dr. A. V. St. George. "Specimen consists of many irregular pieces of friable, gray-yellow tissue designated as a cholesteatoma. Some are partially covered by a white, fibrous layer. One piece of reddish, fibrous tissue, designated as 'capsule,' measuring about 1.5x0.4 cm., accompanies the tumor. *Microscopic:*

(a) (b) (c)

FIG. 7(a).—Case 3: Roentgenogram made after operation. Approximate size of tumor outlined by broken white line. Bony peninsulae over lateral sinus preserved. (b) and (c) Photographs made four and one-half years after operation.

The above capsule reveals it to be a thin fibrous membrane lined by atrophic squamous epithelium. Several friable, irregular pieces of tissue are composed by amorphous acellular material. No cholesterol crystals seen."

It is not known what part of the caseous cholesteatomatous mass was examined for cholesterol crystals. On reexamination of the slide it was observed that no hair follicles or glands were present. The lining membrane, part of which was in apposition to the pericranium, presented the three layers similar to that seen in Case 1.

Postoperative Course.—Uneventful recovery. On the seventh postoperative day the bilateral papilledema was receding. He showed no signs of a cerebral focal nature. He was up and about the ward in two weeks, when he developed a sore throat with elevation of temperature to 103° F., which, however, subsided rapidly. He was discharged, May 21, 1934, 21 days after the operation, with no complaints.

Shortly thereafter he resumed his work and has continued to work to date. He was examined in November, 1938, and the area at the site of operation was sunken rather than bulging (Fig. 7, b and c). His eyegrounds were found to be normal by Dr. Hugh McKeown, but on account of some convergent weakness at near point he was given glasses. Pupils were equal, reaction to light and accommodation was normal, fundi and visual fields were normal.

COMMENT.—Typical diploic type of epidermoid which destroyed first the inner table of the skull, later the outer table in several areas, and left bony islands attached to the pericranium. It was sufficiently large to compress the left cerebellar lobe, lateral sinus and brain stem. His only symptom, headache referred to the occiput and left eye, was probably due to compression of the aqueduct and left lateral sinus. Morsellation preferred to any attempt to remove in toto for the reason that it was mechanically impossible; it also seemed better judgment to subject the patient to a second evacuation of the area rather than to open the dura or risk damage to the lateral sinus. Complete relief.

Case 4.—Bellevue Hospital, History No. R-18: *Epidermoid (cholesteatoma); frontal region; right. Marked erosion and destruction of orbit; proptosis; enucleation of eye. Operation. Recovery.*

V. O., female, Negro, age 46, was admitted to the Eye Service for third time, January 5, 1937, with chief complaint of swelling above the right orbit. Transferred to Neurologic and Neurosurgical Service, January 18, 1937.

Past History.—Negative except for mild diabetes, controlled by diet.

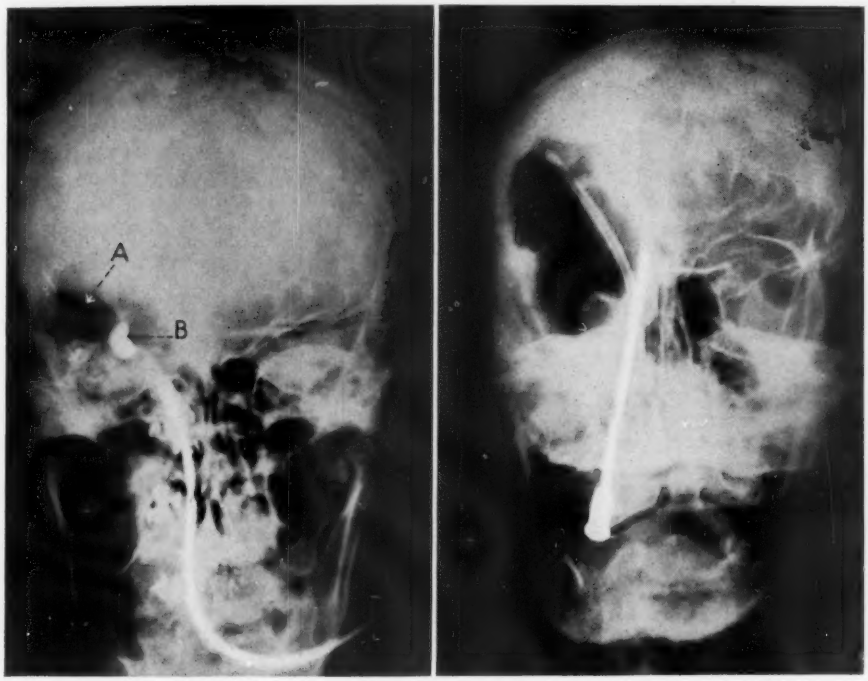
Present Illness.—In 1932, the patient entered the hospital complaining of bulging of, and pain in, the right eye. Enucleation of the eye was performed for suspected tumor. No tumor was found in the orbit to account for proptosis. Two years later she entered the hospital for the purpose of plastic surgery to enable her to wear an artificial eye. She wore the eye until two months before the present admission to the hospital, at which time it became dislodged, fell and broke. This was due to the fact that a swelling had appeared in the lateral half of the right brow about five months ago. It had increased in the last two weeks and extended downward and laterally. The swelling was accompanied by very little pain and only slight headache. There had been no other symptoms, and no loss in weight. Sight in the left eye was good.

Physical Examination revealed a well developed colored woman, height about five feet six inches, weight about 145 pounds. The right frontal sinus did not transilluminate, while the left did. On the right brow and over the outer half of the supra-orbital ridge there was a firm, tense, but slightly compressible tumor, 5x7 cm. in size, which extended downward into the orbit and outward beneath the temporal muscle. It offered resistance similar to that of a firm sebaceous cyst. It was not tender. There was erosion of the upper rim of the orbit and the supra-orbital ridge in its outer half. Neurologic examination was negative in every respect. Blood pressure 140/80. Blood count and urine normal. Wassermann negative. Spinal fluid: Initial pressure 125 Mm. of water, normal dynamics, no cells, no globulin, total protein 40. Visual fields normal in left eye. Right eye had been enucleated. Left fundus normal. Smell test showed normal reaction to citral, with an increased minimal identifiable odor to coffee as well as an increased fatigue for coffee on the right side.

Roentgenologic Examination showed marked destruction of right orbit, anterior ethmoids, the lesser wing of the sphenoid, and, to some extent, the greater wing of the sphenoid, with a cyst-like area revealed in the outer wing on the right side. There was an oval hiatus involving the outer and inner tables of the skull in the right frontal region extending into the temporal region with destruction of the outer half of the supra-orbital ridge, and upward for a distance of about one and one-quarter inches, to a position higher than that of the uppermost limit of the frontal sinus on the opposite side. The left frontal sinus was clear for the most part, but slightly cloudy toward the midline. The margins of the defect were not as clear-cut and defined throughout as it is in the usual case of intradiploic epidermoid. This was probably due to the position of the lesion involving and eroding the thinner bones as it did.

An attempt was made to inject the remnants of the frontal sinus with lipiodol (Fig. 8, a), but only a bead-like mass, about 1 cm. long, was demonstrated, completely filling the remnant of the frontal sinus near the midline. An epidermoid was suspected, but the preoperative diagnosis was not made.

Operation.—January 22, 1937: Avertin and local anesthesia. An incision about two and one-half inches long was made parallel to and about one and one-half inches above the right supra-orbital ridge extending into the temporal region. The flaps consisting of scalp, galea, muscle and pericranium were dissected up, reflected and held with self-



(a) (b)

FIG. 8(a).—Case 4: Roentgenogram showing defect (A) produced by erosion of tumor and lipiodol in remnant of frontal sinus (B). (b) Roentgenogram showing postoperative cranial defect after removal of tumor. A catheter passed through artificial opening made for drainage between cavity occupied by tumor and the right nasal passage. Cannula passed into remnant of frontal sinus.

retaining retractors. The cranial defect, shown on the right side of the frontal bone roentgenologically, could then be made out. Through this defect protruded a semi-fluctuating, encapsulated mass, with a firm fibrous wall, the mass measuring about one inch in horizontal diameter, three-quarters of an inch in vertical diameter, and protruding through the defect about one-half inch above the outer table of the skull. It did not pulsate.

When the capsule was nicked, a bead of thick, viscid, opaque, olive-green material, as viscid and stringy as thick rubber cement, exuded very slowly, like stiff tooth-paste from a tube. When the opening into the cavity was enlarged, a somewhat globular mass of the olive-green, viscid, odorless material was extruded with a slow creeping, rolling movement like lava down a hillside. Its consistency was that of thick coal tar. As more of the material was evacuated, it was observed that the surface of the substance rose and fell rhythmically, synchronous with the pulsation of the brain. More bone was removed until the cranial defect was about 5x7 cm. (Fig. 8, b), the longest diameter directed obliquely backward and upward, extending beneath the temporal

muscle. The thick material from the depths was less homogeneous, did not have the same olive-green color, but was muddy yellowish-green. After this portion of the contents was removed, the material in the deeper part of the cavity was more caseous, like the interior of an intradiploic tumor, and was yellowish-white. The deepest portion of the contents of the cavity, in apposition to the dura, was made up of pearly-white material, similar to that found on the exterior of an intradiploic epidermoid. This layer was about 8 Mm. to 1 cm. in thickness and was plastered against the dura, so that it had to be removed by scraping and scooping it out with a brain spoon.

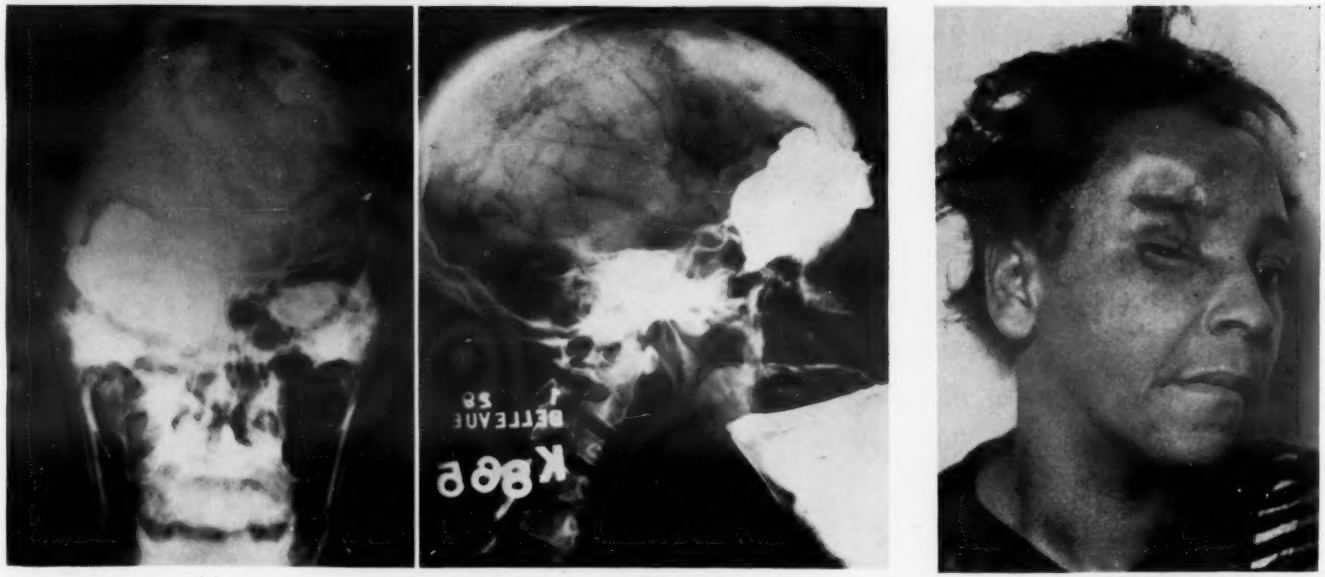
The dura was markedly depressed backward and upward, was concave on the outer surface like the cupped-palm of one's hand, and pulsated. An oval area of dura about 2x2½ ins. was exposed, the inner table being absent. Above, and on the outer side, it was held back by a rim of the inner table of the skull about one-half inch in height with a definite hard, fairly sharp narrow border, similar to the ridge about the internal margin of the bony defect found in Case 1. Otherwise, the inner table was completely destroyed, and the outer table of the frontal bone, where it was not completely destroyed, was considerably and irregularly thinned-out. Once the major portion of the cavity was emptied, two openings, about 1 cm. in diameter, were seen in the antero-inferior wall. These two openings led into several small cavities, incompletely separated by bony septa, as was observed in Case 1 of Munro.[11] These smaller cavities were filled with the same thick, olive-green, viscid material first observed. Nowhere had the cavity broken through into the nasal cavities or antrum, although the anterior ethmoids, the roof and floor of the orbit, the outer half of the supra-orbital ridge, the lesser wing of the sphenoid, a portion of the anterior clinoid process and a portion of the greater wing of the sphenoid had been eroded and destroyed by the slow, increasing compression common to these lesions. The orbital contents could be palpated. The tumor had mechanically accomplished the bony removal advised in the Naffziger[21] operation for progressive exophthalmos better than one could have been effected by operation.

The floor of the anterior fossa had given way before the lesion to such an extent that the lowermost portion of the cavity was delimited by the anterior wall of the sphenoid, later well shown by filling the cavity with lipodol. It was considerably lower than the floor of the sella. This portion of the cavity could be identified on the pre-operative lateral films. The lining of the cavity, where it was firmly fixed to the dura, was yellowish-white in color, and apparently avascular, and did not bleed when scraped fairly vigorously. The dura showed no tendency to fall forward and obliterate the cavity, which was also observed in Case 1. On the contrary, it remained in an arched position, but pulsated in its central portion.

After removal of the bony septa from the antero-inferior portion of the cavity, so that all the cavities had been converted into one, saline solution was poured into the one large cavity, which required about 75 cc. to fill it. None escaped into the nasal passages or throat, showing that no communication existed between the cavity and the nose or nasopharynx. No attempt was made to dissect the membrane from the dura for fear of opening the dura and producing meningitis. The entire cavity was filled with iodoform gauze, the end of which was brought out through the central portion of the incision, and the incision on either side was closed with interrupted silk sutures.

Cholesterol crystals were recovered postoperatively from some of the remaining flakes removed from the cavity. An area of the lining showed stratified squamous epithelium three to six layers thick. There were no hair and no glands.

Postoperative Course.—The patient made an uneventful recovery. On January 29, 1937, the seventh day after operation, and after all the iodoform gauze was removed, the cavity was filled with lipiodol to the level of the skull, the central portion of the incision having been closed with clips to prevent its leakage, and roentgenograms made merely to show the position and extent of the lesion (Fig. 9, a and b). The posterior and superior scalloped borders of the defect were thus made more apparent. One could readily see that the lowermost limits were below the sella and adjacent to the sphenoid



(a) (b) (c)

FIG. 9(a) and (b).—Case 4: Roentgenograms made after filling cavity with lipiodol showing size and extent of the lesion. Anterior bony sphenoid wall apparently eroded. (c) Photograph made November 10, 1938.

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sinus which was almost perforated. There was no leakage into either the nose or mouth.

The cavity was repeatedly packed through the opening in the central portion of the incision and could be inspected readily. The membranous lining of the dura was yellowish-white and did not tend to granulate. The cavity was swabbed out and filled with 20 per cent silver nitrate solution at successive dressings to destroy the remaining portion of the lining epithelial membrane. The walls later changed in color to pinkish-red, became covered with granulations, but the cavity did not fill in rapidly. Nevertheless, later roentgenograms, made after the cavity was filled with lipiodol, showed that it had decreased in size.

Closure of the opening in the scalp over the cavity was desired, but before doing so drainage into the nasal passages was thought to be advisable. A probe was pushed by Doctor Atkinson through the thin intervening tissue between the cavity and the right nasal passage, somewhere in the neighborhood of the frontonasal duct. The tip of a small catheter was tied to the eyelet of the probe and drawn through into the cavity (Fig. 8, b). This did not allow sufficient draining. Therefore, the opening through which the catheter was passed was enlarged to the size of the diameter of a cigarette, and two small rubber tubes were passed through this opening into the cavity, with the proximal ends protruding through the right external naris and fixed with adhesive, which allowed of irrigation with return flow.

Scalp-plastic under local anesthesia was performed; the incision healed kindly and has remained closed. Subsequently one tube was substituted for the two and later still, the patient was able to pass a catheter easily and irrigate the cavity without help. She continued to do this for several weeks, then gave it up, inasmuch as the return flow was always clear.

A photograph (Fig. 9, c) taken in November, 1938, shows the depressed area over the site of the cranial defect and the loss of the right eye. The orbital contents no longer bulge so that she can now again wear an artificial eye. Recent roentgenologic studies following instillation of lipiodol have not been undertaken, but the cavity is doubtless much smaller, if not obliterated. She is perfectly well and does her daily work.

COMMENT.—This case again shows the enormous erosive and destructive tendency of the lesion involving bony structures without gross damage to the soft parts, *i.e.*, dura, brain and orbital contents. It is also noted for the utter lack of neurologic findings. Study of good stereoscopic films, made before any operation was performed, should have warranted the diagnosis of an intracranial lesion with earlier removal of the lesion and preservation of the eye. On account of the loss of the right eye, one could make no deductions regarding the second, third, fourth and sixth cranial nerves, but considering the extent of the bony excavation, one was surprised that the olfactory nerve and the first division of the fifth nerve remained intact.

Rather than to have risked the possibility of intradural infection, no attempt was made to remove the lining membrane completely. Knowing that the cavity was to remain open for quite a while, it was believed that the membrane could be destroyed by a solution of silver nitrate. Should débris collect, it could be washed out through the opening into the nose.

The semisolid, viscid consistency of the major portion of the lesion was probably due to partial "dilution" by secretions of the mucous membrane of the eroded ethmoids and frontal sinus.

Case 5.—New York Hospital, No. 215378: *Epidermoid (cholesteatoma); intradural, middle fossa; left. Marked erosion of petrous, middle fossa and homolateral*

clinoids. Double vision for 12 years. Involvement of left cranial nerves III, IV, V, VI, VII, and VIII. Operation. Recovery.

J. S., white, female, single, age 40, was admitted to the hospital, November 1, 1938, with chief complaint of double vision for 12 years.

Family History.—Father had ulcers of stomach. Mother had tuberculosis. Both in fair health. One sister well. Two brothers; one had tuberculosis. Family nervous and high strung.

Past History.—At age of six fell and struck head. Not able to "talk straight" for a while. Pneumonia as child. Tonsils removed at 16. Appendicectomy at 20. Otherwise general health excellent until beginning of present illness.

Present Illness.—Began in 1926. At time an impacted left wisdom tooth and an impacted incisor removed; noticed numbness left side face and forehead. Numbness never improved. Intermittent double vision began about this time, worse during warm weather and under nervous tension and worry.

In 1927, right impacted wisdom tooth removed, operation on right antrum, and bone removed from nose. During 1929, double vision became continuous; vision of left eye blocked by dark covering. A number of eminent ophthalmologists could not find cause or remedy. Glasses prescribed in 1932. Operation for double vision considered but not undertaken. May 31, 1934, bilateral myringotomy by Dr. Ross Faulkner; profuse discharge from both ears, culture showing *Streptococcus hemolyticus.* Pain and discharge continued. June 12, 1934, bilateral mastoidectomy performed by Doctor Faulkner. Mastoids filled with pus and granulation. Discharged June 23, 1934. Office dressings. Both wounds healed by July 11, 1934. After operation diplopia disappeared for six months.

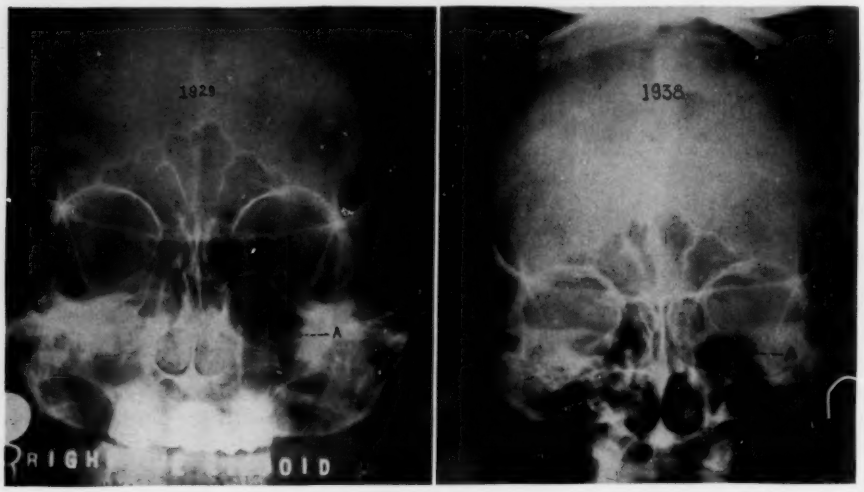
In the period between 1936 and 1938, had several corneal ulcers in the left eye for which she was treated by two capable ophthalmologists. One had observed her since 1934. In 1935, he noticed a partial left facial paralysis which was attributed to the mastoidectomy. He observed several recurrences of corneal ulceration, the last being noted May 5, 1938. Although cornea was anesthetic, complained of pain referable to left eye. Muscular condition unchanged. Interpreted findings as congenital paralysis of superior oblique and external rectus muscles and partial facial paralysis following mastoidectomy with resultant corneal ulceration.

Another ophthalmologist first examined her September 27, 1938. Two infected corneal abrasions of left eye. Right pupil reacted normally to light and accommodation, left pupil fixed and somewhat dilated. Corneal sensation normal right eye, left cornea anesthetic. Marked limitation motion of left eye in all directions; third, fourth and sixth nerves involved. Vision 20/15 right eye, 20/50 left, with correction. Right visual field normal, marked contraction left central field, with enlarged blind spot. Under mydriasis no evidence of increased intracranial pressure, media clear, disk, blood vessels and macular region left eye normal. From nerve involvement he suspected intracranial pathology.

Physical Examination.—The only positive findings were those referable to left eye and left cranial nerves. Appendicectomy and bilateral mastoidectomy scars. Patient well nourished and not ill.

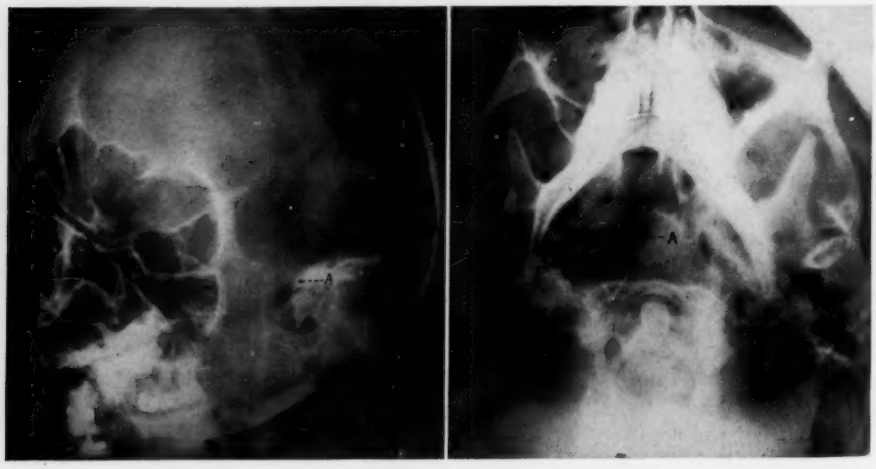
Roentgenologic Examination.—October 18, 1938: Dr. F. M. Law. Stereoscopic films revealed marked erosion of left petrous amounting almost to amputation through its midportion (Figs. 10, b, and 11, a). The eminential arcuata remained, but the trigeminal impression was obliterated. Marked erosion, almost obliteration, of left anterior and posterior clinoids. Size of sella unchanged. Floor of the middle fossa was depressed. No change in appearance of left optic foramen. The stereovertexmental films (Fig. 11, b) showed erosion of the left side of body of sphenoid and marked erosion of the floor of the middle fossa. No outline of left foramen ovale. Apparent fracture at posterior border of left sphenoid. Squama of the left temporal bone thinned-out, especially in lower limits. Reexamination of previous roentgenograms made elsewhere, in 1929 (Fig. 10, a), nine

years previously, also revealed marked erosion of the left petrous, similar to that shown in the more recent films. In the latter, however, the major extension of the erosion was shown to have involved principally the lower portion of the petrous. *Roentgenologic Impression:* Meningioma of the middle fossa.



(a) (b)

FIG. 10(a).—Case 5: Roentgenogram, made in 1929, showing destruction of petrous tip (A) by erosion of epidermoid of middle fossa. (b) Destruction of petrous (A) only slightly increased after nine years; roentgenogram made in 1938.



(a) (b)

FIG. 11(a).—Case 5: Oblique roentgenogram showing erosion of petrous (A) in 1938. (b) Vertexmental roentgenogram showing erosion and partial destruction of sphenoid bone (A). Left foramen ovale not seen.

Neurologic Examination.—Dr. Foster Kennedy: "Double vision since 1926, off and on. Persistent since 1934. Occasional headaches. Daily pain in left eye and left face. Pain comes on gradually and recedes gradually. Headache in back of head 'at the base of skull.' Worse at the end of day. Never wakes her out of sleep. No difficulty in use of arms or legs. No pains down arms. Hearing in left ear impaired since 1927. Lost

double vision for a few months after operation (mastoidectomy) in 1934. Has gained seven pounds this summer. Now weighs 115 pounds.

"Fundi show congestion, but no frank papilledema. Left 3rd, 5th, 6th and 7th nerve involvement with slow progression over 12 years. Left motor 5th nerve with atrophy of temporal and masseter muscles. Marked diminution of left corneal response. Weakness of left 7th nerve peripheral. Impaired hearing, almost deafness, left ear. Left petrous ridge like a 'bridge blown up in middle.' Otherwise neurologic examination is negative. Operation advised."

Operation.—November 2, 1938: *Craniotomy for tumor of left middle fossa.* Avertin, local and ether anesthesia. Following the advice of Adson,[20] the left external carotid artery was first ligated to control bleeding. It was thought that the lesion probably was a meningioma of the middle fossa, of saddle-bag type. Curved incision made through scalp and galea; flap reflected forward. A low flap consisting of temporal fascia, muscle, pericranium and bone was turned down. Squamous portion of temporal bone was very thin in its lower limits and could be bent like celluloid, and was absent over an area about 1½ cm. in diameter. Dura appeared normal. Before opening dura, a mass, the shape of the small end of a hen's egg, could be palpated low in the temporal region projecting from the middle fossa, extending outward toward the squamous portion of the temporal bone, posterior to the sphenoidal ridge, in the typical position of a middle fossa meningioma which extends to the surface. Dura opened low and surface of lesion exposed. Sphenoidal portion of the temporosphenoidal lobe was displaced upward and backward by lesion. When the overhanging flange of the sphenoidal portion of the lobe was elevated, more of the tumor came into view. It had a distinct, firm capsule or wall, grayish-white in color, and about the same texture, resiliency and toughness of the dura itself. The presenting portion of the tumor was not adherent to dura or undersurface of brain. It did not have the appearance of a meningioma. It was softer and seemed partially fluctuant. No material obtained on aspiration. Through a nick in the most prominent apical portion of the lesion, small amount of oily material exuded. When incision was enlarged typical mother-of-pearl cholesteatomatous material, resembling that usually found in an epidermoid, was encountered. The covering of the visible portion of the tumor was removed in a circular manner, just as one would remove the top of a soft-boiled egg. Several sutures were placed in the remaining cut margin of the wall, and the capsule was split downward in several places in order to enlarge the opening into the encapsulated mass. The contents were scooped out with a brain spoon and suction, until the cavity was empty. The weight of the material removed was 60 Gm. (Fig. 12, a).

The inner lining of the capsule was pinkish-white in color and did not bleed, but oozed slightly. With the brain spoon, the resistance encountered was similar to that offered by a half-ripe alligator pear. The wall or capsule was not adherent in its outer half, either to the dura or brain surface, but was very adherent to the structures forming the floor of the middle fossa, over the remaining portion of the left petrus, and to the dura in the low temporal region. Erosion of the petrous, floor of the middle fossa, posterior and anterior clinoid processes, and the lateral surface of the body of the sphenoid was marked. In the eroded areas of the bone there were undulations amounting at certain places to small pockets. Into these pockets the cheesy material was snugly packed.

Lying in the bottom of the cavity one could see the internal carotid artery, from the point where it makes a right-angled turn in the petrous, forward to the origin of the anterior cerebral artery. The posterior half was denuded and completely exposed, and one had fear lest it might become weakened and develop an aneurysm in this portion. Its outer wall was cherry-red. It was not covered by dura. From the point where it emerged from the petrous forward it was covered with dura. The internal carotid artery was palpated so that no mistake could be made about its identity. Its branches could be seen clearly through the thin covering. The petrous tip and the superior border with a considerable portion of the anterior and posterior surfaces had entirely disappeared.

The tumor was roughly the shape and size of a duck's egg, with the larger end

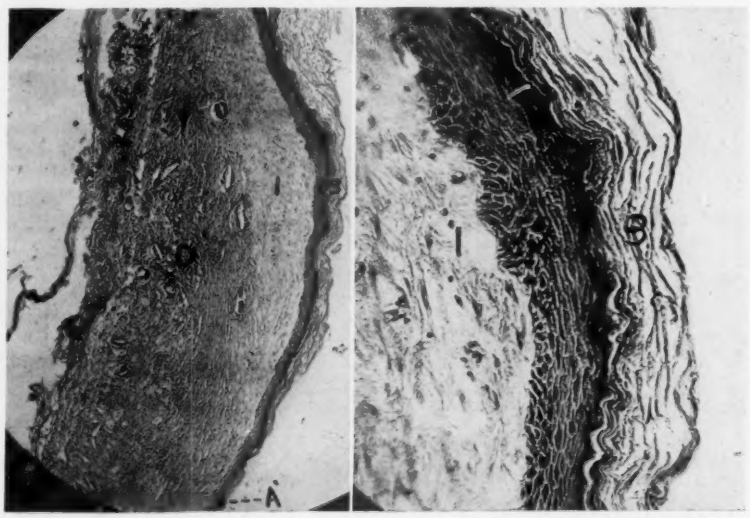
(a) (b)

FIG. 12(a).—Case 5: Fragments of intradural epidermoid occupying middle fossa. (b) Cheesy fragments of epidermoid wrapped in cellophane and placed *in situ* in left middle fossa to show approximate position of tumor.

flattened against the structures described and packed in the middle fossa (Fig. 12, b). It was about three inches deep and more than two inches in diameter in its largest portion. A considerable part of the capsule or wall was removed, but no attempt was made to remove it from the depths, realizing well that there might be partial, slow refilling of the cavity. Should this occur, the contents could be removed through a short, straight incision, as in the operation for major trigeminal neuralgia.

The fifth, sixth, fourth and third nerves were not identified, although the position of the gasserian ganglion was seen. It was believed that once the enormous, though slow, pressure was released from these structures by removal of the contents of the epidermoid, function through these nerves would be restored, providing they were not impregnated with cholesterol crystals.

The cavity was allowed to collapse as much as it would, the distorted sphenoidal lobe was allowed to drop back over it, and the dura was closed except in the lowermost



(a) (b)

FIG. 13.—Case 5: Photomicrographs of wall of epidermoid showing the three layers (1, 2 and 3). (a) Low power, showing partial destruction of epithelial layer from pressure (A). (b) High power, showing 10 to 12 layers of epithelial cells.

portion. A small gauze wick was placed through the dural incision in order to further collapse of the cavity, to allow escape of cholesterol containing fluid, and to control slight oozing. This strip of gauze was brought out through a puncture opening in the middle of the pedicle of the muscle-fascia flap and through the transverse portion of the incision above the zygoma. The flap was returned to its position, wired in three places with rustless steel wire, and the scalp incision was closed in two layers. A transfusion was given on the table by Doctor Eggston.

Pathologic Examination.—Gross: Dr. N. Chandler Foot. "Specimen consists in part of a roughly triangular piece of silvery-white membrane measuring 1.5 cm. on the side. This is apparently composed of connective tissue resembling dura, and at one point there is a small mass of whitish cheesy material similar to that to be described later, adhering to it. The greater part of this specimen consists of 47 Gm. of whitish-yellow soft material which has the consistency of pot cheese. (Some of the tumor was lost.) The largest fragment measures approximately 3 cm. in diameter, but most of it is broken into flaky white curds which contain nothing resembling true tissue. *Microscopic examination* shows that the bulk of material is nothing more than masses of keratinized, exfoliated

horny layers from the epidermis. Sections made from the small sheet of tissue (Fig. 13, a and b) show it to consist of a layer of stratified squamous epithelium, which is divisible into a basal layer, prickle cell layer, stratum granulosum and stratum corneum, which shows marked overproduction of keratinized material. This epidermoid tissue extends along a part of the membrane only and tapers off to be replaced by granulation

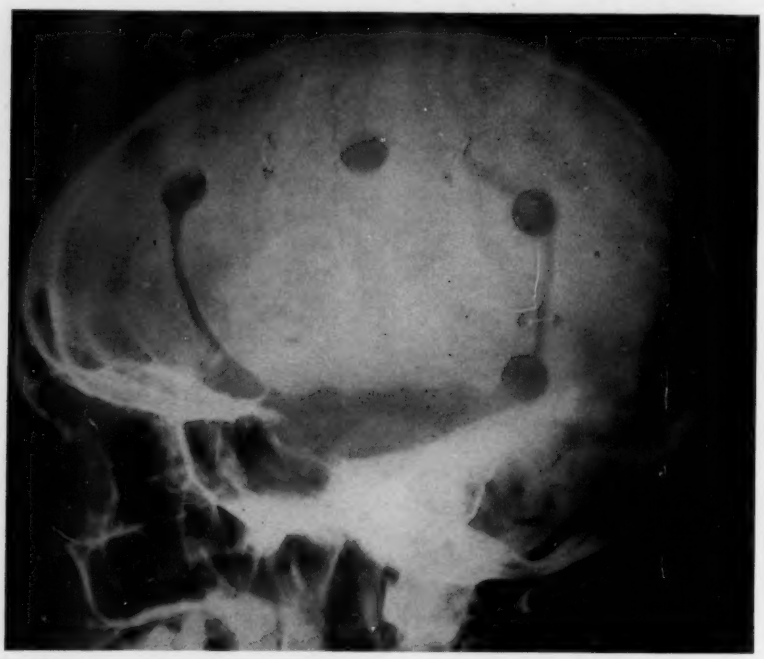


FIG. 14.—Case 5: Roentgenogram made after operation showing removal of thin squama, fixation of bone flap with rustless steel wire, and erosion of left anterior and posterior clinoids.

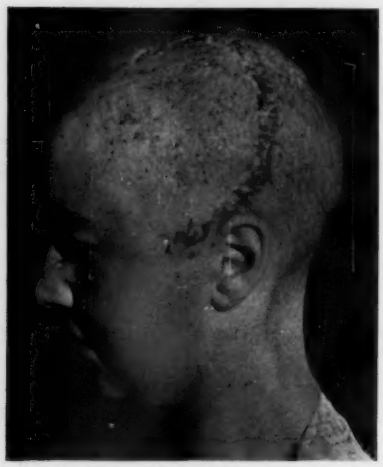


FIG. 15.—Photograph 12 days after operation, showing position of craniotomy scar in the hair line and the transverse cervical scar of incision through which ligation of external carotid artery was accomplished.

tissue in which there are large numbers of foreign body giant cells, many containing acicular crystal spaces. There are very large numbers of fat phagocytes containing innumerable small, clear vacuoles which give them the typical 'foam cell' appearance. Underlying this layer of epidermal tissue there are no accessory organs such as sebaceous glands, sweat glands or hair follicles, proving this to be epidermoid rather than dermoid in nature. *Pathologic Diagnosis:* Epidermoid (cholesteatoma) of brain."

Postoperative Course.—The patient made an uneventful recovery. There was at first slight difficulty in word finding (anomia) which cleared up before her discharge. The gauze wick was partly removed on the first, and completely removed on the second day. This was accompanied by a considerable amount of bloody, watery fluid escape with the pulse beat. Both incisions healed *per primam* (Figs. 14 and 15). On the second post-operative day she was mentally

clear. Out of bed on tenth day. She was discharged, November 18, 1938, 15 days after the operation.

Neurologic Examination.—Pupils normal except left, slightly irregular; right fundus normal. Left disk showed an area of pallor without engorgement of veins or swelling of disk. Visual fields grossly normal. There was complete paralysis of left sixth nerve with difficulty in upward lateral gaze to the left. Corneal reflex absent on left. Incomplete left fifth nerve paralysis. Definite partial paralysis of seventh nerve. Almost total deafness on left. Other cranial nerves normal. Sensation normal over rest of body. No astereognosis or adiodokokenesis. Knee and ankle jerks active and equal. Abdominal reflexes present and equal. Plantar responses bilaterally flexor. No asynergy. No difficulty in speech. Motor functions normal. Walks well.

COMMENT.—The slow progress of these lesions is especially exemplified in this case. For three years there was no neurologic sign or symptom other than intermittent diplopia, the left eye being involved. For about nine years diplopia was constant except for six months' interval following mastoidectomy in 1934. Slow involvement of other cranial nerves followed, caused by a lesion evidently involving the left middle fossa. Roentgenograms made nine years before operation showed marked erosion of the petrous tip and other bony structures forming the walls of the middle fossa. Doubtless, had roentgenologic examination been made several years prior to 1929, they would also have shown erosion. Over 12 years intervened before the diagnosis was established. Diagnosis probably should have been made. Given a case with similar history, roentgenographic and neurologic findings, one *should* make the diagnosis, enabling one to remove the major portion of the lesion through a smaller opening in the skull low in the temporal region without making an osteoplastic flap. Involvement of the nerves should probably clear up either partially or wholly unless the nerve trunks have become impregnated with cholesterol crystals.

Case 6.—Hospital for the Ruptured and Crippled. Hosp. No. 825–307: *Epidermoid (cholesteatoma); intraspinal, fourth and fifth lumbar. Widening of canal. Corroboration with lipoidol. Hemilaminectomy. Recovery.*

D. C., white, male, age 17, single, first admitted to the hospital, July 8, 1932, complaining of pain in small of back and both thighs, stating that onset of pain was sudden about two years previous to admission. At that time the pain was knife-like in character and lasted about two days. It continued for several weeks as a dull ache. It occurred whether lying, walking or sitting.

Physical Examination disclosed a somewhat awkward gait, without limp. Kyphosis extended from the cervical VII to lumbar IV; exaggerated deep reflexes, and no sensory changes except hypersensitiveness to pain, especially in the sole of right foot. Gave history of "meningitis" in 1926, with apparently good recovery and freedom from symptoms until onset of pain in 1930. Not stated whether lumbar punctures were done during the course of the meningitis.

Lumbar puncture performed, August 17, 1932—only 5 cc. fluid obtained, with great difficulty. Pressure on jugular caused slight increase in the flow of fluid. Diagnosis of chronic adhesive leptomeningitis or adhesive arachnoiditis made. Examination under ether revealed definite shortening of the right hamstring muscles. On August 29, 1932, the hamstrings were lengthened. Previous to this, with the thigh flexed on the abdomen, the knee could not be extended more than 150 degrees. On September 6, 1932, a plaster spica was applied under avertin and ethylene anesthesia, with the right lower extremity at right angles with the trunk.

Neurologic Examination.—October 20, 1932: After the spica was removed, there was right foot-drop, complete loss of motion in foot, some atrophy of calf muscles, and trophic areas on right heel and metatarsal areas and sole of the right foot. Sensory impairment was present in the distribution of the internal and external popliteal nerves, most pronounced in the external. Diagnosis of pressure neuritis (pressure from spica) was made. Improvement of motion in foot followed removal of spica but no appreciable improvement in sensation. Ulcerated areas on sole of foot finally healed, but the ulcer on heel remained. Discharged, November 26, 1932, to return to O. P. D.

Admitted again, February 2, 1934, for ulceration of right heel. Atrophy of right thigh was noted. Discharged, May 8, 1934.

Third admission, September 21, 1934. Lumbar puncture performed, October 1, 1934, at fourth lumbar space. No fluid obtained. At third lumbar interspace, pressure of fluid was 10 Mm. of mercury, and on pressure of jugular vein elevation to 23 Mm. of mercury. After this admission, he developed retention of urine which was quite disturbing, requiring catheterization.

Neurologic Examination, October 8, 1934, showed atrophy of the entire right lower extremity with swelling of the distal parts, duskiness of the skin; areas of old pressure necrosis on heel and beneath metatarsal healed. Practically complete paralysis of muscles of right leg and foot. Could neither extend nor flex foot. There was anesthesia in distribution of external popliteal, hyperesthesia in area supplied by internal popliteal and posterior tibial, right knee jerk exaggerated, left normal, no Babinski, marked exaggeration of right gluteal reflex, pain on deep pressure over lower lumbar region and urinary retention. Lumbar laminectomy advised.

The author saw the patient in consultation later, and corroborated the above neurologic finding that there was complete motor paralysis of the muscles of the right leg and foot. The patient could weakly extend and flex the leg. There was definite hypesthesia and anesthesia in the area supplied by the external popliteal nerve. The area of hypesthesia extended just above the midpoint of the leg on its outer surface, and anesthesia was present in the lower portion of the leg and dorsum of the foot. The plantar surface of the right foot was more hyperesthetic than the left. There was no saddle hypesthesia or anesthesia. Right knee jerk greater than left. Ankle jerk absent on right. No Babinski. Gluteal reflex on right markedly exaggerated. Whole lower extremity on right showed atrophy. Pressure over lumbar IV and V vertebrae produced pain. Inspection of ordinary roentgenogram showed increase in width of the interspinal space, especially at L IV and V. It was believed that a lesion involving the cauda equina was present. Lumbar puncture at fourth interspace was advised. Partial block might thus be determined or a complete lack of fluid would demonstrate presence of an occluding lesion. Lipiodol injection then advisable. Lumbar puncture done following day and 2 cc. lipiodol injected. Roentgenograms showed the lower point of the neck of the lipiodol medial to the body of the fourth lumbar vertebra, and there was a small streak extending slightly lower on left side. Twenty-four hours later it was at same level. It capped the lesion with its upper limit at the level of the upper border of the body of the fourth lumbar vertebra. Right hemilaminectomy advised.

Operation.—December 3, 1934: *Hemilaminectomy (as advised by Dr. Alfred S. Taylor), with removal of the right laminae of L III, IV and V.*

The laminae were thinned-out. When the dura was opened, the upper rounded but slightly pointed pole of an encapsulated tumor was seen. Its color was grayish-white and in appearance it was quite similar to the lining membrane of the lesion observed in Case 5. The presenting pole was the shape of the small end of a robin's egg, the larger end being toward the sacral portion of the canal. The lesion was partly covered with the component parts of the cauda equina. The nerve roots making up the cauda equina were highly injected and pinkish-red in color. This was thought to be due to the lipiodol which had been in the spinal canal since October 9, 1934. The lipiodol had changed in color and consistency from a clear, homogeneous yellow oil to a yellowish-white, opaque,

oily substance. A number of globules of lipiodol had become encysted, some along the exit of the roots of the cauda equina and some along the roots themselves. The major portion was free in the subarachnoid space and could be removed. The tumor had the consistency of a typical epidermoid with a wall or capsule about 1 Mm. thick. No fluid was obtained from the lesion on aspiration. The upper pole was removed and a section of the limiting membrane preserved for examination. Grayish-white, cheesy, crumbly material was removed. It had the typical appearance of epidermoid or cholesteatomatous débris. The contents of the sac were scooped out with a dull curet. The lesion was one and one-half inches in length and about three-quarters of an inch in its widest portion. The interior of the sac was pinkish-white. Some of the material adjacent to the inner surface of the limiting membrane had the pearly-white luster observed in the other case.

After the contents of the sac had been removed, the cavity collapsed partially. Although the upper pole of the lesion was free and nonadherent, the greater portion of the remainder of the lining membrane was intimately adherent to the component parts of the cauda equina and could not possible have been removed without grave damage to the nerve roots. It was believed better judgment would be exercised in leaving most of the capsule rather than to attempt to dissect it away from the nerve roots, especially on account of the encapsulated, encysted bodies of lipiodol already found involving the nerve roots above. Therefore, the lamina of the first sacral overlying the lower portion of the lesion was not removed. Cerebrospinal fluid flowed into the cavity which remained after partial removal of the wall. The dura was tightly closed.

Microscopic Examination of a section of the lining membrane revealed that it consisted of three layers very similar to the lining membrane in Case 1, 2, 3 and 5. The contents of the tumor showed many cholesterol crystals. No hair follicle or glands were seen. *Pathologic Diagnosis:* Cholesteatoma (epidermoid).

Postoperative Course.—Uneventful recovery. The bladder paralysis soon cleared up, general condition improved, and he began to gain weight. Right foot-drop with paralysis of the entire foot persisted. January 11, 1935: Calcaneocuboid arthrodesis and posterior bone block performed by Dr. Marvin Stevens, to stabilize the foot. He was discharged, February 15, 1934, and received outpatient treatment in the Orthopedic Clinic. The trophic ulcers of the heel and metatarsal region recurred after patient was up and walking, and, on February 8, 1937, he was admitted for the fourth time for further treatment of the ulcerated areas. Ulcers healed, and he was discharged, March 14, 1934. On January 19, 1938, he was admitted to the hospital for the fifth time for consideration of sympathectomy, in an endeavor to heal the ulcerated areas in the right foot. This procedure was abandoned after a study of the skin temperatures. Roentgenologic studies of the spine after lipiodol injection again revealed definite block in the region of the third lumbar vertebra. Another exploration of the spinal canal was advised.

Operation.—February 2, 1938: *Hemilaminectomy, right.*

Exposure was made as at the previous operation with widening and extension of the exposure below. Some bone had formed at the site from which the fifth lamina had been removed; otherwise, the dura was covered with scar tissue. The dura was tense and, when excised, a considerable amount of clear fluid escaped with several small cholesteatomatous masses. It was not known whether this débris had accumulated since the last operation, or was material which had not been removed at that time. At any rate, the amount was small. The original tumor cavity had become sealed over at the upper pole so that it had no communication with the subarachnoid space. The lipiodol recently injected above did not escape through the incision. In order to evacuate it, a probe was passed upward through the adhesions and this opening was enlarged by stretching. Lipiodol escaped together with a considerable amount of cerebrospinal fluid. The opening was made larger with the hope that it would remain patent, and the subarachnoid space would be continuous with that of the epidermoid sac. The roots of the nerves making up the cauda equina, although compressed, were not so reddened and irritated as they had been when observed at the first laminectomy. Incidentally, it was observed that the

lumbar region was quite strong following the original hemilaminectomy, due to retention of the spinous processes, intraspinous ligament, and laminae of the opposite side. The area from which the laminae had been removed was filled with strong fibrous tissue. The exposure was quite sufficient. The dura and incision were closed.

Since paralysis of the external popliteal nerve following the previous tenotomy and stretching of the hamstring muscles persisted, and also in view of the fact that there was practically complete loss of sensation in the foot, it was considered advisable to explore the external popliteal nerve at the level of the tenotomy incision opposite the knee joint to determine if the nerve had been injured. There was no division either of the internal or external popliteal nerves found, nor was there any marked amount of scar tissue along the sciatic nerve at its bifurcation or about the external or internal popliteal nerves. All three nerves, however, appeared to be quite small and atrophic, the atrophy being most marked in the external popliteal.

Postoperative Course.—Uneventful recovery; patient sitting up in bed on the fourteenth day with the wound healed. Two days later, up in a wheel chair. The trophic ulcers were completely healed. On March 1, 1938, the area in the metatarsal region was excised by Doctor Huber. The great toe was amputated and the skin of the great toe turned under to cover the defect. The flap healed well. Patient was up and about the ward in 12 days, bearing weight. On April 2, 1938, the skin flap, which at first was in excellent condition, had partly broken down. Weight-bearing withheld and the area healed. Anesthesia of dorsum of foot persisted. Patient allowed up and walked well. General condition good.

COMMENT.—This is another instance in which the diagnosis of the intraspinal epidermoid was delayed for several years after the first operation. Due to delay in recognition and removal of the tumor, the patient was admitted to the hospital five times, was given many anesthesias, and underwent six operations. He was in the hospital 429 days, from July 8, 1932, to May 20, 1938. He also has a crippled right lower extremity, which doubtless is permanent. It is possible that complete removal of the tumor with its lining membrane might have been accomplished on his first admission.

It is not known whether the paralysis of the external popliteal and partial paralysis of the internal popliteal nerve were due to overstretching under anesthesia, or compression of nerve by the spica or whether this was due to the intraspinal tumor. It is possible that the roots of the nerves forming the cauda equina may have become infiltrated with cholesterol crystals as was the fifth nerve in the case described by Mahoney.[14]

On account of the encysted condition of the lipiodol found at the first laminectomy, it is believed laminectomy should be performed following injection of lipiodol, as soon as the level of the lesion can be determined. Taylor's hemilaminectomy suffices, and is advisable for removal of tumors in the region of the cauda equina.

Case 7.—Bellevue Hospital, No. K.—774(26): *Epidermoid (cholesteatoma); posterior fossa; anterior to pons and medulla, extending through foramen magnum. Operation: partial removal of tumor. Death.*

Y. E., white, female, housewife, age 28, admitted to Neurologic and Neurosurgical Service, September 27, 1929.

Family and Past History.—Essentially negative.

Present Illness.—Suffered with generalized headaches for six years. Headaches be-

came occipital in location two years before admission. Became more severe six months before admission, associated with double vision and pain in the eyes.

Neurologic Examination.—Coarse nystagmus on left lateral gaze and fine nystagmus on right lateral gaze. No upward nystagmus. Fundi normal. Visual fields normal. Right lower facial weakness. Diminished sensation over left face with diminished left corneal reflex. Palate moved more toward the right side. Hoarseness of voice probably caused by left vocal cord paralysis. Atrophy of left sternomastoid and trapezius muscles. Atrophy left side tongue. Pressure tenderness over left suboccipital region. Relative weakness right arm and leg. Succession movements slightly awkward with right hand. Deep reflexes slightly increased on right. Abdominal reflexes diminished on right. Right plantar equivocal, left plantar flexor.

T.P.R. normal. Serology normal. Roentgenologic examination of skull negative. Spinal tap revealed initial pressure of 240 Mm. of water, with *almost complete block. Preoperative Diagnosis:* Extramedullary neoplasm, lying to the left of and ventral to the medulla.

Operation.—September 27, 1929: Dr. Alfred S. Taylor. *Combined suboccipital exploration and high cervical laminectomy.*

A midline incision from the external occipital protuberance to the fourth cervical spinous process was made. Bone from suboccipital region, laminae and spinous processes of C I and II removed. Dura overlying cerebellar lobe, medulla and cord very tense; did not pulsate. No palpable tumor through dura before opening. Posterior horn tapped to reduce intraventricular pressure. When dura was opened, definite pressure collar about cerebellar lobes and tonsils, evidently caused by jamming into foramen at time of lumbar puncture. Cerebellar lobes otherwise normal. Medulla appeared broadened and on palpation thought to be hard. Hardness found to be due to neoplasm which displaced the medulla backward and to the right. Tumor glistening, encapsulated, firm, nodular and grayish-white. Tumor lay anterior and to left of medulla, with stretching and atrophy of cranial nerves where they emerged from left side of medulla. Extent of tumor upward beneath medulla and pons not determined. Length, therefore, not known. Only partly removed. Considerable hemorrhage throughout. Closure. Left table in very poor condition and died shortly arriving on ward. No autopsy.

Pathologic Examination.—"Cholesteatoma." No further report made, but microscopic examination showed a stratified squamous epithelial lining with epithelial débris; no hair or glands.

COMMENT.—Slow development of signs and symptoms shown again in this case. No bony erosion noted. Tumor apparently occupied a position from which it could not be removed. Death probably due, or at least hastened, by preoperative lumbar puncture, with jamming of the conus.

Case 8.—Bellevue Hospital, Hosp. No. M.—242: *Epidermoid (cholesteatoma);* right frontal lobe. *Operation: Osteoplastic flap with tapping of cholesteatomatous cyst. Reelevation of flap and removal of tumor. Recovery.*

E. B., white, male, age 33, was admitted to the Psychopathic Division and transferred to the Neurologic and Neurosurgical Service, July 18, 1931; the first of three admissions, with complaint of nocturnal convulsions.

Family History.—No convulsions in family. One brother mentally deficient. Wife without pregnancy.

Past History.—Always healthy, except for scarlet fever. No operations or accidents, except dislocation of shoulder during convulsions.

Present Illness.—Ten years before admission, onset of convulsive seizures; all with exception of two occurred while asleep. At first occurred about three times a year, gradually increased in frequency until three months before admission when convulsions occurred about twice a week. They lasted from one to three minutes. Characterized by general

convulsion, not originating in jacksonian fashion. Did not bite tongue but usually lost vesical sphincter control. Recalled nothing about attacks except that he fell during one and dislocated his left shoulder. Did not know if he lost consciousness, and there were no bad after-effects. Two years before admission there was complete loss of libido and potency, and for one year had progressive impairment of memory.

Physical Examination.—Well developed male, not ill, overcooperative and unusually pleasant. Some weakness of left shoulder thought to be due to previous dislocation. General physical examination negative.

Neurologic Examination.—Essentially normal with exception of bilateral, equally diminished weak tendon reflexes, but thought to be within physiologic limits. Slight right facial weakness. Absent abdominals on right, sluggish on left. Disk margins hazy. *Clinical Diagnosis:* Idiopathic epilepsy with grand mal seizures. Unlocalized frontal lobe tumor to be excluded.

Subsequent Course.—Three encephalograms were made resulting in some, but not sufficient, evidence to warrant operation. Serology normal. Initial spinal fluid pressure at each of the lumbar punctures for the purpose of making encephalograms about 200 Mm. of water, with normal dynamics and normal findings in the fluid. Patient discharged, September 15, 1931, as unimproved.

Second admission December 16, 1931, complaining that convulsions had become more frequent, but still without noticeable after-effects. Neurologic findings showed increased tendency to anosmia, increased deterioration of memory, loss of libido and potency, frequent recurrent convulsions as described, equivocal weakness of right lower face and left arm. Normal reflexes, except for abdominals, which were absent except in upper left quadrant. Cremasterics absent on right. Fulness of veins and hazy disk margins on right.

On December 28, 1931, ventriculograms were made by Dr. John E. Scarff. Bilateral bur holes over posterior horns. Right ventricle not located. Thirty cubic centimeters of cerebrospinal fluid were removed from the posterior horn and replaced with air. *Roentgenologic examination* showed deformity of anterior horn of right lateral ventricle, and diagnosis of right frontal lobe tumor was made. The patient was much improved following ventricular puncture until, three weeks later, he became violent and was returned to the psychopathic ward with restraints. Operation for removal of tumor of right frontal lobe was further deferred because of an infection of the thigh and septic temperature, which later cleared up.

Operation.—January 27, 1932: Dr. Byron Stookey. *Frontotemporal craniotomy, with evacuation of cholesteatomatous cyst.*

Avertin, local, gas-oxygen and ether anesthesia. The dura was tense. Exploratory puncture made with cannula through right frontal lobe in direction of ventricle. At a depth of 1.5 to 2 cm. a cyst was encountered and 90 to 100 cc. of colorless fluid were removed. This fluid was filled with white flakes which quickly settled when allowed to stand. Flaky particles had gross consistency of soft paraffin or cheese. Microscopic examination showed cholesterol crystals. Diagnosis of cholesteatoma with cystic formation was made. Transcortical incision in cyst was considered but deferred on account of patient's poor preoperative general condition. Closure.

Postoperative Course.—Following evacuation of cyst, the patient ran a stormy course with bronchopneumonia and evidences of meningitis. Temperature to 105° F., for which repeated spinal taps were made with intraspinal administration of antimeningococcic serum. Cerebrospinal fluid was slightly cloudy and xanthochromic, 330 Mm. of water initial pressure, final 160 Mm. No block. Cell content, 4,400 cells, mostly polys. Negative culture. Wound healed without infection. After a stormy course, condition improved sufficiently to allow of elevation of flap and extirpation of tumor.

Second Operation.—April 21, 1932: Dr. Byron Stookey. *Reelevation of bone flap with removal of tumor.*

Local anesthesia. Cortex overlying lesion appeared normal. Incised with electric

cautery. Surface of tumor encountered at depth of 1 cm. "Typical cholesteatoma, composed of irregular flakes of waxy material, of very soft consistency and easily broken up by pressure." About 60 cc. of material removed with large scoop. Capsule of tumor exposed. It appeared tough and well defined. Adjacent brain carefully brushed away from capsule. Thoroughly done except at one point where a small elongation of capsule dipped down into the depths and appeared to have definite attachment. This was amputated low, and cauterized. Elsewhere removal of capsule complete. Practically no bleeding. Cavity in brain filled with saline. Dura tightly closed. Replacement of flap.

Pathologic Examination.—Gross: "Specimen consists of a mass of yellowish, friable tissue broken into small pieces. Mass after fixation weighed 65.3 Gm. *Microscopic.*—Lining membrane: Section showed tissue comprising wall of cyst composed of several rows of stratified epithelial cells, with a small amount of keratin on the surface. In the wall of the cyst are numbers of fairly large cells with abundant cytoplasm containing small nuclei, of epithelial origin. *Pathologic Diagnosis:* Epithelial lining of cholesteatoma." (Epidermoid.) No hair or glands seen.

Postoperative Course.—Uneventful recovery and rapid improvement. Discharged, June 7, 1932, to return to follow-up clinic.

Third admission, November 13, 1933, on account of persistence of convulsions even under strenuous luminal therapy. Readmitted for complete check-up. Convulsions became less, and duration of each attack shorter. Continued absence of libido and potency with progressive mental impairment. General condition otherwise good.

Neurologic Examination.—Right anosmia, left central facial weakness, grip stronger on right, slight adiodokokenesis of left arm. Gait normal and no Romberg. Abdominal reflexes, right greater than left. Fair average intelligence to test. No marked personality defect. There was an occasional tendency to be overbearing and surly. Spinal tap showed normal initial pressure with normal dynamics and fluid. Discharged, November 18, 1933.

COMMENT.—This case also shows marked slowness in the development of the lesion. It is the only case of the series in which the major portion of the tumor was *within* the brain substance. The origin of the tumor was not known. It was not intraventricular. It probably arose from some of the structures situated anteriorly adjacent to the skull, or near the midline, and pushed upward into the brain. One might deduce this from the fact that the small elongation of the capsule "dipped down deeply and appeared to have a definite attachment." It is possible that the irritative meningitis, with marked increase of cells, but sterile cultures, might have been produced by leakage of the fluid containing cholesterol, as described by Mahoney.

Of the eight cases reported (Table II) six are personal and two were operated upon by other surgeons (Doctors Taylor and Stookey). Five were male, and three female. The average age was 31. The youngest was 19 and the oldest 46. Therefore, the lesion was found more frequently in the third and fourth decades. All came to operation. Seven recovered, and one died. Preoperative diagnosis was made in two instances (Cases 1 and 3), and should have been made in two others (Cases 2 and 4), and probably in Case 5. The first known preoperative diagnosis was made in Case 1.

SUMMARY

(1) A brief review of the literature has been given, including a discussion of the nomenclature. The terms "epidermoid, extradural diploic" and "epi-

TABLE II

CASES OF EPIDERMOID TUMORS (CHOLESTEATOMATA) REPORTED IN THIS SERIES

Case No.	Sex	Age	Operator	Interval Between Onset and Operation	Chief Neurologic Findings	1. Palpable Tumor 2. Size and Weight 3. Location	Removal	Result
1 M. D.	M.	28	King	5 yrs.	Weakness left leg, blurred vision, weak convergence, left hypesthesia, increased reflexes on left, slight dizziness and mental impairment	1. Yes 2. 110 Gm. 7x5x4 cm. 3. Diploic, right frontoparietotemporal	Complete	Recovery
2 J. R.	M.	27	King	Not known	None	1. Yes 2. About 22 Gm. and 2 cm. diameter 3. Diploic, left frontal	Complete	Recovery
3 B. M.	M.	26	King	1 yr.	Constant headache for 1 yr. Occipital pains radiating to left eye and right occipital region	1. Yes 2. 110 Gm. 3. Diploic, left occipital	Incomplete removal of lining membrane	Recovery
4 V. O.	F.	46	King	3 yrs.	Marked protrusion of right eye, and double vision. Otherwise none	1. Yes 2. Estimated 100 Gm. 3. Diploic, right frontal	Incomplete removal of lining membrane	Recovery
5 J. S.	F.	40	King	12 yrs.	Diplopia, paralysis or paresis left 3, 4, 5, 6, 7, 8 cranial nerves. Mild headaches	1. No 2. 60 Gm. 3. Left middle fossa	Incomplete removal of lining membrane	Recovery
6 D. C.	M.	19	King	4 yrs.	Low back pain; pain, weakness, atrophy and sensory disturbances right lower extremity, anesthesia dorsum of foot, right foot-drop. Retention of urine	1. No 2. Estimated 30 Gm. 3. Intraspinal, lumbar 4 and 5	Incomplete removal of lining membrane	Recovery
7 Y. E.	F.	28	Taylor	6 yrs.	Coarse nystagmus to left. Fifth nerve paresis, left, right seventh, left 10, 11, 12 paresis, weakness right arm and leg. Spinal block. Pressure tenderness right occ.	1. No 2. Not known 3. Anterior to medulla, extended through foramen	Specimen removed	Died (no autopsy)
8 E. B.	M.	33	Stookey	10 yrs.	Essentially normal. Absent abdominals, right, sluggish on left. Disk margins hazy	1. No 2. 65.3 Gm. 3. Right frontal lobe	Complete, except piece of small deep elongation of wall. Coagulated	Recovery

dermoid, intradural" are suggested. These terms designate the origin and location of the lesion, and the confusing term "cholesteatoma" is avoided. The only reasons for retention of this term are that it readily indicates the nature of the contents of the lesion and also because of its established place in the literature.

(2) Epidermoid tumors are relatively rare, the intradural type being about four or five times more frequently observed than the extradural type. More cases have been reported in the last two decades than have ever previously been reported. Prior to 1920, the tumor was revealed first at autopsy in the majority of cases.

(3) The living portion of the tumors is the covering membrane, of which the epithelial layer only is active. They are slow in growth but relentless in their eventual destructive force. On account of the benign nature of the lesion, early recognition and removal is indicated.

(4) Points in the roentgenographic findings in these lesions which produce bone erosion have been discussed. Most of these cases should be diagnosed and in some instances completely removed.

(5) Eight cases of epidermoid, all of which were operated upon, are reported. Seven recovered and one died.

The author is indebted to Dr. Alfred S. Taylor and Dr. Byron Stookey for permission to report Cases 7 and 8, respectively.

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DISCUSSION.—DR. GILBERT HORRAX (Boston, Mass.): Although Doctor King has covered the subject exhaustively, there are a few points which I would like to discuss concerning these rare and unusual types of intracranial lesions. In an earlier communication, I used the term cholesteatoma. One must not misunderstand that. Certain types of cholesteatoma are the collections of débris found in the middle ear, and this term has likewise been used for the craniopharyngiomata. These have no relation to the tumors which Doctor King has been discussing. In addition to the epidermoids, there are intracranial dermoids which evolve from a deeper layer of inclusion. It is particularly with the dermoid variety that I wish to deal, because Doctor King has covered the epidermoids so fully. The dermoids represent a very low percentage of all intracranial tumors, perhaps not more than 0.3 per cent. For some reason they may become infected even before operation, and then one must deal with them along the same lines as an abscess.

I have had two that became infected. One was in a child who had a suboccipital fistula before operation was undertaken. The other point about these tumors is that they may involve any layer from the scalp to the intracranial cavity, subdural or in the diploe. One large intradural cholesteatoma, which I reported some years ago, was in a patient in an insane asylum.

She has made an excellent recovery since the tumor was removed. They are all potentially benign tumors. There is one question which has been raised, and that is as to how much one should do with these tumors, that is, how much to take out. Sometimes it is possible to get the whole tumor if it lies fairly superficially, but some of these in the cerebellopontile angle may run far forward into the middle fossa and, in my opinion, it is almost impossible to get all the material out. I have one such patient who has been well for ten years since operation.

Dr. Walter E. Dandy (Baltimore, Md.): Doctor King's German statistics indicate that almost half of these tumors are located alongside the pons and in the cerebellopontile angle. This, too, has been my experience. This is one very important reason for employing the cerebellar route in operating for the cure of trigeminal neuralgia. Over 5 per cent of all cases of trigeminal neuralgia are caused by tumors in this angle, and the pearly body tumors comprise about half of these. By the cerebellar operation, the tumor can be removed in addition to curing the tic pain; thus two birds are killed with one stone. If the temporal approach is employed, all of them will be missed. The pearly tumors are not large, are very slow growing, are bloodless and easily removed.

THE TREATMENT OF INTERNAL CAROTID ANEURYSMS WITHIN THE CAVERNOUS SINUS AND THE CRANIAL CHAMBER

REPORT OF THREE CASES

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THE surgical attack upon intracranial aneurysms is just beginning. Nor is it the insuperable problem that the desperate nature of the lesions might lead one to believe. Indeed, a few have unquestionably been cured. Within the past few years several aneurysms have been disclosed at operation, others demonstrated just as unequivocally by Moniz's arteriography, and procedures of different types have been directed toward their cure. In general, the surgical attack has been of two kinds—(1) direct, and (2) indirect. The former attempts to deal with the aneurysm directly, the latter indirectly by inducing thrombosis: (1) Through ligation of the internal carotid artery—Hunter's method of proximal ligation; or (2) by trapping the aneurysm between ligatures. The three aneurysms included in this report have been treated by the latter method. The type of operation will always be dictated by the position and to some degree by the size of the aneurysm.

Although the direct attack upon aneurysms would appear to be the more logical and more certain of permanent results, it is only certain aneurysms that are amenable to such treatment and, at best, the procedure must be regarded as one carrying definite hazards. Two successful direct attacks upon intracranial aneurysms have been recorded, both appearing during 1937, and each was operated upon by a different method. McConnell[11] (1937) incised an aneurysm (probably from the internal carotid artery) and despite a furious hemorrhage plugged the sac with muscle. The visual defects resulting from the aneurysm have since disappeared, and the patient is free of symptoms and is back at work.

The writer[4] (1937) closed a silver clip upon the neck of an aneurysm of the internal carotid artery and shriveled the aneurysm distal to the clip with the electrocautery. The clip was flush with the wall of the carotid. The third nerve palsy cleared completely in six weeks and the patient is perfectly well and free of all symptoms, 21 months later (December, 1938).

Dott[5] (1937) placed silver clips upon both sides of an aneurysm of the anterior communicating artery but an unfortunate postoperative accident deprived him of a beautiful result—the patient died from an intrathoracic puncture of an infusion needle.

Tönnis[20] (1936) by splitting the corpus callosum, made a most difficult exposure of a cherry-sized aneurysm of the anterior communicating artery (shown by arteriography), found the aneurysm surrounded by a "walnut-sized" hematoma. Fearing injury to the arterial trunk from which the aneurysm arose, he merely covered its surface with a piece of muscle. The

patient made an uneventful recovery. However, one cannot believe that muscle applied to the outer surface of the aneurysmal sac could play any part in curing the aneurysm; possibly it might, as the operator hoped, seal the leak and prevent further bleeding. Dott[5] (1933) wrapped a sheath of muscle around an intracranial aneurysm of the carotid at the junction with the posterior cerebral artery and later[5] (1937) reported the patient well after four years. Here again, one cannot believe the application of muscle could have had any curative effect upon the aneurysm. Trevani[21] (1932) evacuated an intracranial carotid aneurysm, suspecting it to be a tumor, when aspiration was negative; the internal carotid artery was ligated in the neck to control bleeding, but the patient died five hours later. Sosman and Vogt[18] (1926) included in their paper on the roentgenologic examinations of aneurysms of the circle of Willis, a case operated upon by Cushing; an aneurysmal sac on the intracranial portion of the carotid had been deliberately opened and was packed with muscle. The patient, age 58, recovered with hemiplegia, doubtless due to closure of the carotid artery. There was partial return of motor function but she died six months later. A note from Doctor Sosman related that at necropsy the aneurysm was found to be thrombosed.

By the indirect attack, there is reason to believe that several intracranial aneurysms have been cured. Dott, who has just presented a most interesting series of intracranial aneurysms, is probably entitled to the credit of the first successful surgical treatment by simply ligating the internal carotid artery. His first cure was reported in 1933, and is doubtless the one that was reported well four years later (1937). In his later report (1937) there is good reason to believe that three of his verified aneurysms have been cured by simply ligating the internal carotid artery in the neck. The remaining four of his seven verified cases died, although two were unconscious when the operation was performed. Walsh and Love[22] (1937) reported another almost certain cure by the same method of ligating the internal carotid in the neck. Subsequent photographs showed the oculomotor palsy to have disappeared and the patient was back at work three months after the operation. In Jefferson's[10] series of carotid-cavernous aneurysms (1938) is one (Case 5) demonstrated by angiography and probably cured by this method. Another (Case 15) developed transient contralateral motor numbness 36 hours after ligation of the internal carotid, but at the time of his publication was reported to be improving.

In the accompanying report three additional cases of intracranial aneurysms are added. All arose in the intracavernous portion of the internal carotid artery or just as the carotid enters the cranial chamber. All projected into the cranial chamber alongside the carotid artery. In each there is every reason to believe that the aneurysm is cured. The method of treatment has consisted of trapping the aneurysms between two occlusions of the internal carotid artery (1) intracranially; and (2) in the neck. This method is similar to that carried out by the writer in the treatment of carotid-cavernous-arteriovenous aneurysms.

CASE REPORTS

Case 1.—History No. U-73513: L. B., white, female, age 28. Admitted September 25, 1936, discharged October 15, 1936. Referred by Dr. Roger G. Doughty and Dr. J. Heyward Gibbes, Columbia, S. C.

Complaints.—Headache and drooping of the left eye.

Family and Past Histories were negative.

Present Illness.—Four or five months ago generalized headaches appeared, became periodic, but were not severe. Six weeks ago, sudden, intense headache appeared over the left side of the head and continued for 24 hours. After the headache had subsided the vision was impaired, but she did not then see double. Objects seemed a little less clear than previously, but she could not say definitely that only the left eye was affected. Four days later there was another similar, equally severe attack that lasted for two days and again the pain was mainly over the left side of the head. She was not exactly unconscious but she could remember very little that happened during that period. For two or three days before this attack there was mental haziness. Patient was taken to a hospital and repeated lumbar punctures done; blood was obtained from these punctures. These, she thought, gave her temporary relief.

A third and similar attack of pain occurred three weeks ago. At this time the left eyelid drooped and the eyeball pulled outward. Since that time she has been unable to open her left eye. There has since been a constant dull headache with exacerbations on three different occasions.

Physical Examination.—Patient is somewhat undernourished; Blood pressure, 130/70; Wassermann, negative.

Neurologic Examination showed complete paralysis of all motor functions referable to the left third nerve. Patient was unable to elevate the eyelid even slightly. The pupil was widely dilated and did not react to light or accommodation. There was also complete paralysis of the left fourth nerve; the sixth was intact. The following negative findings are perhaps worthy of note: Both visual fields were normal; the visual acuity was not recorded. The eyegrounds were normal on both sides. No murmur was audible over the head. Compression of the left carotid artery over a period of five minutes produced no symptoms. Roentgenologic examination of the head was negative. *Clinical Diagnosis.*—Aneurysm of the left carotid artery or the posterior communicating artery.

Operation.—October 6, 1936: A hypophyseal flap was turned down on the left side, using the concealed incision. Behind the left carotid artery was a small, rounded, reddish mass, about the size of a pea, bulging from the inner margin of the dura lining the middle fossa of the skull, *i.e.*, covering the cavernous sinus (Fig. 1). When this little mass was touched with the forceps it pulsated strongly. It appeared to be part of the carotid as it passed through the dura. When the carotid artery was gently separated from the aneurysm with forceps there was a small spurt of arterial blood (Fig. 1 inset). This ceased as soon as the carotid fell back upon the arterial wall; this gentle separation was repeated three times and with each there was a spurt of arterial blood. In other words, the posterior wall of the carotid artery was blocking the small opening in the aneurysm and this was doubtless the source of bleeding on former occasions. The third nerve could be seen pushed backward and outward by the aneurysm.

A silver clip was placed upon the internal carotid artery and compressed, completely occluding it (Figs. 1 and 4). After closing the cranial wound, the internal carotid artery was then exposed in the neck. This vessel was elevated upon a tape and compressed with the finger. An opening was made into the carotid cephalad to the point of compression; a strong spurt of blood indicated the rapidity with which a return circulation develops from the ophthalmic artery (its principal source of collateral). A small piece of muscle was introduced into the opening of the carotid artery (Brooks' method) and a clip placed upon it for subsequent identification roentgenologically. The artery was then ligated above and below the opening. Roentgenograms subsequently showed the position of the

clip on the piece of muscle to have remained at the site at which it was introduced into the artery (Fig. 4).

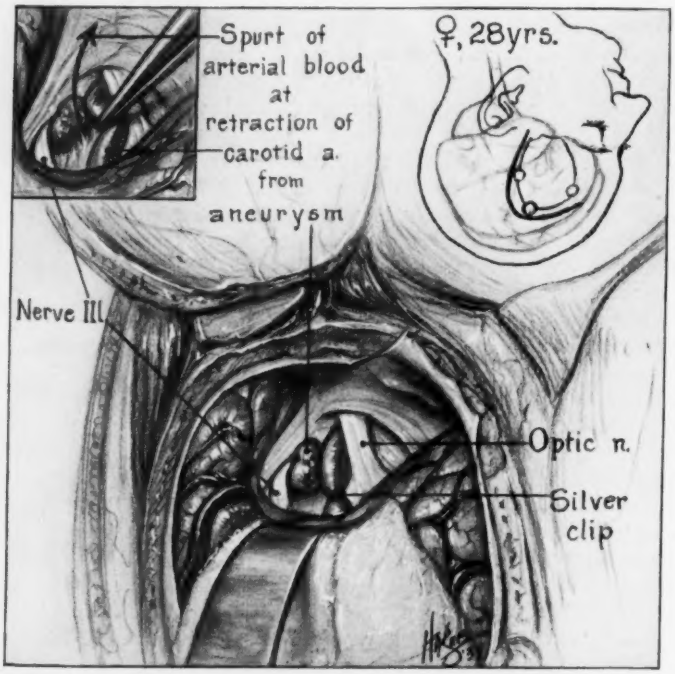

FIG. 1.—Drawing of aneurysm of carotid artery exposed at operation (Case 1). A clip has been placed upon the internal carotid artery intracranially. The upper left inset shows the separation of the artery and the aneurysm by forceps and indicates a spurt of blood that immediately followed the separation. The right upper inset indicates the position and relative size of the concealed cutaneous incision and the bone flap.

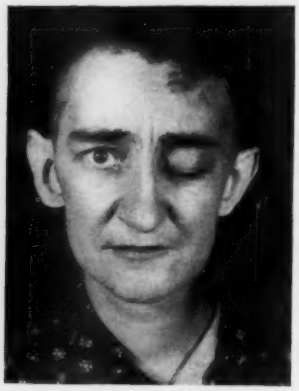

FIG. 2.—Patient (Case 1) before operation. There is complete ptosis of the left upper eyelid.

FIG. 3.—Same patient (Case 1) two years after the operation.

The patient made an uneventful recovery and was discharged October 15, 1936, nine days after the operation.

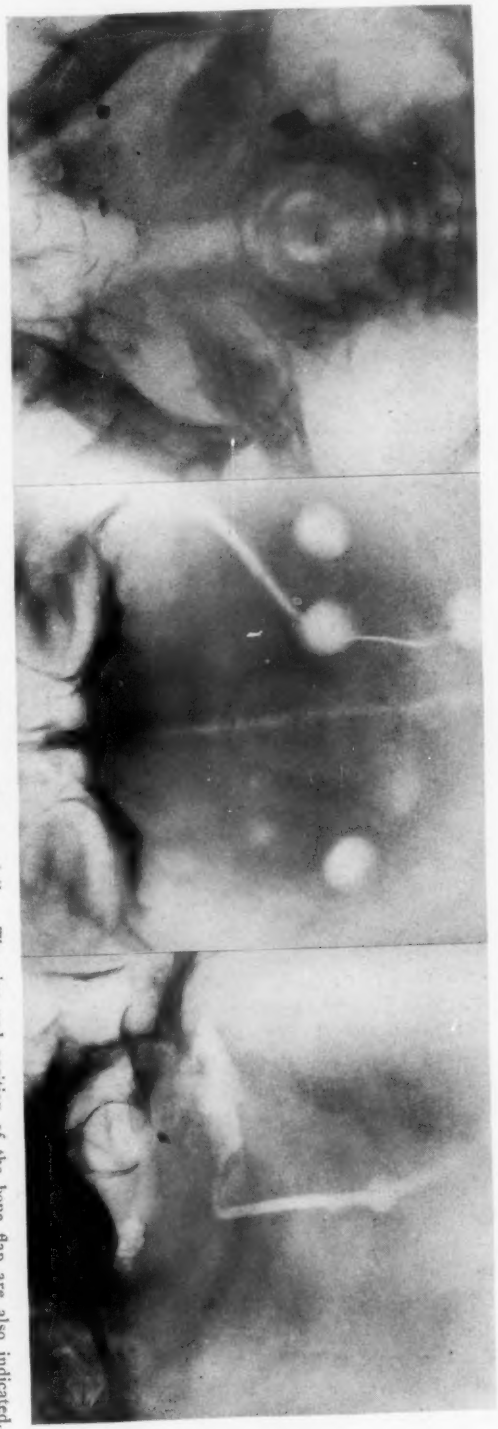

FIG. 4.—Roentgenograms (Case 1) showing the silver clip placed upon the carotid artery, intracranially. The size and position of the bone flap are also indicated.

Subsequent Course.—Patient returned for observation April 6, 1938, 18 months after the operation (Fig. 3). She had been perfectly well; had had no headaches and had been doing all of her own house work. She had gained 32 pounds in weight. A later note, November 15, 1938 (25 months after the operation), says that she still remains well and free of symptoms.

When examined April 6, 1938, the left pupil did not react either to light or accommodation, nor did it react consensually. There was a normal movement inward of the left eyeball, but an almost complete inability to look up or down. The left palpebral fissure was only 2 Mm. narrower than the right, and she could elevate the lid. There was no vision in the left eye, normal in the right. Unfortunately, visual acuity was not recorded before the operation, but the visual fields were normal. There must, therefore, have been a very pronounced loss of vision in the left eye as a result of the arterial ligation, or, more probably, as a result of thrombus formation that included the ophthalmic artery. The left disk showed marked primary optic atrophy and the arteries were greatly narrowed.

COMMENT.—The headaches in this patient have been entirely relieved over a period of 25 months. There has been improvement in the extra-ocular movements, principally the inner movement of the eye, and a marked improvement in the ptosis, but the functions of the extra-ocular muscles have not been completely restored. One is curious to know the cause of the loss of vision in the left eye. The natural assumption is that it is due to the ligature of the carotid, of which the ophthalmic artery is the only accessible branch. However, the same ligatures have been placed on the internal carotid artery in the other two cases, and in several other cases for carotid-cavernous aneurysms, and without loss of vision. It is conceivable, indeed it is my belief, that the small piece of muscle that was introduced into the artery may have led to the formation of a propagating thrombus that occluded the mouth of the ophthalmic artery and perhaps extended along it to the retinal artery. Unfortunately, our records are not sufficiently clear on the preoperative state of vision to warrant any conclusion, though I think there cannot be any doubt that her vision was quite good, although perhaps not perfect, before the operation was performed; certainly her visual field was normal in this eye.

Case 2.—History No. U-137770: C. L. S., white, male, age 36. Admitted April 20, 1938; discharged May 7, 1938. Referred by Dr. Edward F. Milan, of Baltimore, Md.

Complaints.—Headache and trouble with left eye.

Family and Past Histories were negative.

Present Illness.—This began eight months ago with headaches localized to the left orbit and the left frontal region; and associated with these was a tickling sensation in the nose. There was some nasal discharge and he thought he had a fresh cold. Headaches were not constant; they occurred almost every morning and frequently awakened him from a sound sleep. The attacks came on suddenly and without warning, "like a flash," and lasted about one hour; he had one or two every day. If an attack did not come in the morning, it would be certain to come in the afternoon and usually around four o'clock. Since the onset there had never been a day without an attack. They left him exhausted and he had found nothing to give relief.

Five months ago the left eye began to close and draw. This progressed so that he could no longer keep the eye open. Vision in the left eye has been blurred during the last month. At times the ptosis came on in spells lasting from 30 to 60 minutes, and then in large part cleared, but always with a little residual droop. Generally he has

felt "tired and droopy," but there has been no generalized weakness. He has had no convulsions, and has never been comatose. The eye has never protruded. There has been no noise in the ear or head.

Physical Examination was entirely negative. Patient is a well nourished, average-sized man of 36 years. Blood pressure was 120/70, and the Wassermann reaction from the blood was negative.

Neurologic Examination.—Incomplete ptosis of the left upper lid (Fig. 6). Some swelling of both upper and lower lids. Exophthalmometer measured 16 Mm. on the left and 14 Mm. on the right. Exophthalmos, if real, could not be detected on inspection. There was limitation in adduction and downward and upward movements of the left eye. The left pupil was larger than the right but reacted well. Vision in the left

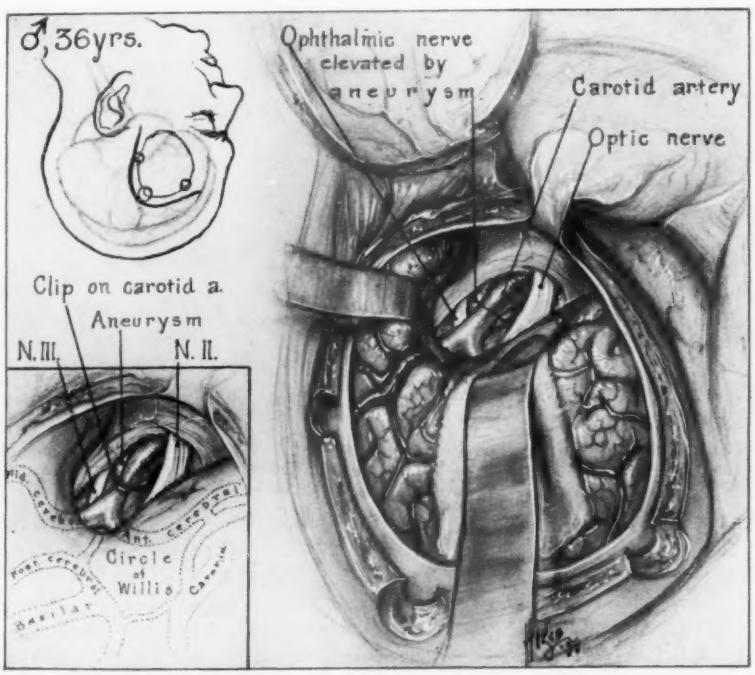

FIG. 5.—Drawing of aneurysm (Case 2), which lies just behind the carotid artery and pushes the oculomotor nerve backward. The difference in color of the carotid artery at the point of entrance into the skull and that farther along on the artery is noted and probably carries some significance. The artery is also larger at the point of entrance into the skull than where it approaches the brain. The lower inset shows the clip placed upon the carotid artery.

eye was 20/30, and the right 20/20; the left could not be improved by lenses. The visual fields were normal; the blind sports were normal in both eyes. No murmur could be heard with a stethoscope. Roentgenologic examination of the head was negative. *Clinical Impressions.*—This patient was first seen in the Diagnostic Clinic, where Doctor Ford made the diagnosis of a tumor, or more probably an aneurysm of the carotid or posterior communicating arteries. My own diagnosis was the same; I thought it was almost certainly an aneurysm.

Operation.—April 22, 1938: A hypophyseal approach was made on the left side, using the concealed incision. This exposure provided ample room for inspection of the carotid artery. A branch of the sylvian vein crossing to the dura along the lesser wing of the sphenoid was thrombosed and divided, and this permitted inspection of the floor of the middle fossa of the skull. The third nerve was seen to be elevated by a lesion beneath,

i.e., from the region of the cavernous sinus. When the optic nerves and the carotid artery were separated one could see a small reddish-brown mass projecting between them on the mesial side (Fig. 5). The region of the cavernous sinus was much fuller than normal and pulsated when the forceps pushed upon it. The branches of the carotid along the base of the brain were visible and the lesion was not attached to any of these. One point was perhaps noteworthy, *i.e.*, in the first half of the carotid intracranially the wall of the artery was pink (Fig. 5), but beginning with a very sharp line about its midpoint the normally white thickened wall continued onward to the base of the brain; it was assumed that the whiter coating meant a greater thickness of the artery. A flat silver clip was clamped upon the internal carotid artery completely obliterating it. (Fig. 5, inset.) This was accomplished without difficulty and without bleeding. Five days later the internal carotid artery was ligated in the neck. The artery was distinctly smaller than usual.

FIG. 6.—Patient (Case 2) before operation, showing moderate degree of ptosis.

FIG. 7.—Front view of patient (Case 2) six weeks postoperative. The ptosis has completely disappeared. In the side view, the cranial scar is seen under the hair line.

Subsequent Course.—Patient returned June 4, 1938, one month after discharge, and six weeks after the first operation. He was perfectly well in every way. His ptosis and all of the extra-ocular palsies had entirely cleared (Fig. 7). He had no pain, no headache and the tickling sensation in the left side of the nose had disappeared. There was no sign of atrophy of the disk. His visual fields were normal on rough test. He said the vision in that eye was perfectly normal. He was seen again August 1, 1938; was perfectly well and was back at his work—a clerical position. He remains well, November 15, 1938.

Case 3.—History No. U-135470: D. R., white, female, age 37. Admitted April 4, 1938, discharged April 30, 1938. Referred by Dr. John F. Daly, Teaneck, N. J.

Complaints.—Drooping of the right eyelid and headaches.

Family and Past Histories were negative.

Present Illness.—For the past five years, patient has been having brief, mild headaches about once a week and readily relieved by aspirin. Four months ago she had a peculiar pain about the right eye; this lasted three days when it became suddenly very much more severe and remained concentrated in the right eye. She described the sensation as though the eye was being torn out. During this attack she fell unconscious. After two or three minutes she was able to get up and went to bed unassisted. A terrific headache and pain over the entire right side of the face persisted. She was nauseated and vomited frequently. At that time drooping of the right upper eyelid and double vision developed. A week later there was a similar attack, following which the right eyelid was completely closed and she was unable to elevate it. A week later she had a third attack. She was then taken to a hospital where she remained 12 weeks.

The physicians said they could hear a "swishing" noise in the head, but the patient has never heard it. When she had obtained no relief during a month's stay in the hospital, the right internal carotid artery was ligated. The physicians told her that the bruit had disappeared, but patient recognized no subjective improvement.

During the past month she has had a noise in the right ear like escaping steam from a locomotive. She thinks there may have been some protrusion of the right eye which has since disappeared; she was, however, not certain of this. Her right pupil has been persistently dilated.

Physical Examination.—Patient is a large, obese woman, seemingly normal, except for her local deformity. Blood pressure 138/96; Blood Wassermann reaction, negative.

Neurologic Examination showed the right eye to be strongly abducted. The pupil was dilated to 6 Mm. and did not react to light either directly or consensually, or to accommodation. All movements of the muscles supplied by the third nerve were completely paralyzed. Slight rotary movement of the eyeball was possible, doubtless due to an intact fourth nerve. The external rectus muscle functioned normally. Ptosis was complete.

Vision in the right eye was 20/50, in the left 20/15. It was impossible to chart the visual field in the right eye because of the very strong abduction of the eyeball. The

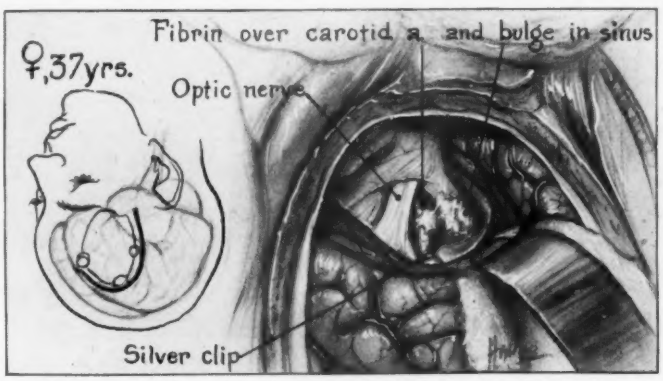

FIG. 8.—Case III: Operative findings: A clip has been placed upon the carotid artery. The layer of fibrin, which was so conspicuous in this case, is undoubtedly an indication of the weakness of the aneurysm.

eyegrounds were normal. The tension of the eyeball was normal on both sides. There was no exophthalmos and no murmur could be heard with the stethoscope. Compression of the right internal carotid artery for five minutes caused no cerebral disturbance. Roentgenologic examination of the head was negative.

Operation.—April 13, 1938: A right hypophyseal approach was made, using the concealed incision. The brain was quite tight and surprisingly little room could be obtained by evacuating the cisterna chiasmatis. The lateral ventricle was tapped and 25 cc. of fluid obtained; even then it was difficult to get sufficient room to expose the carotid artery. In order to gain access to the anterior part of the middle fossa, it was necessary to thrombose and divide two branches of the sylvian vein crossing to the lesser wing of the sphenoid; finally, an adequate exposure of the carotid region was obtained. There was a small piece of fibrin over the outer surface of the carotid artery as it passed through the base of the skull. (Fig. 8). This was cautiously divided and one could see beneath it a bulging pulsating mass, about the size of a pea, extending from the cavernous sinus, which bulged markedly and pulsated (Fig. 8). The small intracranial protrusion was everywhere smooth and glistening and had no relationship to the posterior communicating artery. The third nerve could not be seen because the bulging mass covered its course. The carotid artery was isolated from the optic nerve, a silver clip placed upon it and compressed. There was no difficulty in doing this and no bleeding resulted.

Second Operation.—April 22, 1938: Ligation of the Right Internal Carotid Artery in the Neck:

In another hospital, ligation of the internal carotid artery was said to have been done. The wound had become infected and drained for several weeks, so that there was now an exceedingly dense scar throughout the depths of the old incision. Cautiously the scar tissue was cut away until the carotid artery was located. The common carotid artery was finally exposed, then the internal and external carotids for a short distance. Both the internal and external carotid arteries were essentially normal in size and both pulsated normally. We could find no ligatures or fascia or foreign material of any type that had been placed around the carotid. The internal carotid artery was then ligated with a double ligature of medium silk; a second ligature was placed just above the first one. On palpating the vessel, cephalad to the ligature, no pulsation could be detected. The patient made an uninterrupted recovery and was discharged April 30, 1938 (Fig. 9).

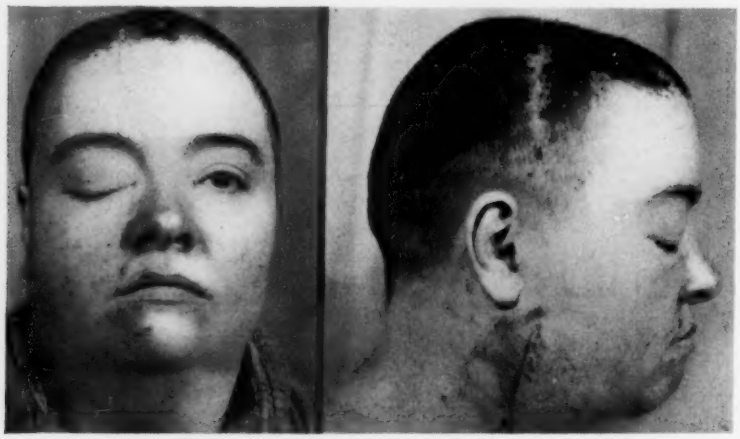

FIG. 9.—Front and side views of patient (Case 3) immediately after operation. The complete ptosis is precisely the same as before operation.

Subsequent Course.—Patient wrote, July 10, 1938, that the ptosis was improving; she could elevate it slightly. She returned for observation, August 27, 1938; she could then elevate the lid almost to the normal, though when at rest it lagged markedly (Fig. 10). There was just a beginning abduction of the eyeball; the upward and downward movements were unimproved and the pupil though less dilated than formerly did not react to light. Oblique rotation of the eyeball was present. The vision and visual fields on the affected side were normal. The vision in the affected eye was 20/20; before operation it was 20/50. Her general health was perfect. She had had no headaches and the noise in the head had disappeared. In a later photograph (Fig. 11), taken in December, 1938, eight months after the operation, a still further improvement is noticeable.

Diagnosis of Intracranial Aneurysms.—The intensive cultivation of any field usually discloses the lesion to be of far greater frequency than we have been wont to believe. This has already been found to be true with intracranial aneurysms. Sporadically, cases have been reported for over half a century. Blane (1800) and Adams[1] (1869) reported specimens at necropsy. Hutchinson[9] (1875) presented a postmortem report of one that had been diagnosed correctly 11 years earlier. A spontaneous cure had resulted from thrombosis. During the past decade the number of cases has increased quite

rapidly; in most instances there are several from a single clinic. Jefferson[10] has just reported 17 aneurysms in the cavernous sinus alone, and in his experience has had 55 arterial aneurysms of the brain; many of these, however, have not been verified either by operation or necropsy.

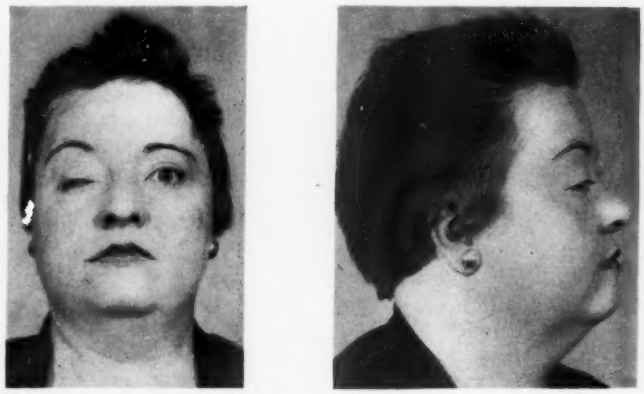

FIG. 10.—Front and side views of patient (Case 3) four months after the operation. The ptosis is beginning to clear.

The diagnosis of an intracranial aneurysm may be made with the greatest of ease or with great difficulty. Similarly, the localization of the aneurysm, which, of course, is all essential for surgical treatment, may be simple or difficult. Unfortunately, the easy localizations from clinical signs and symptoms are in the minority. However, arteriography, introduced a decade ago by Moniz,[16] frequently makes the diagnosis and localization with precision, when the signs and symptoms fail completely, or are at least uncertain.

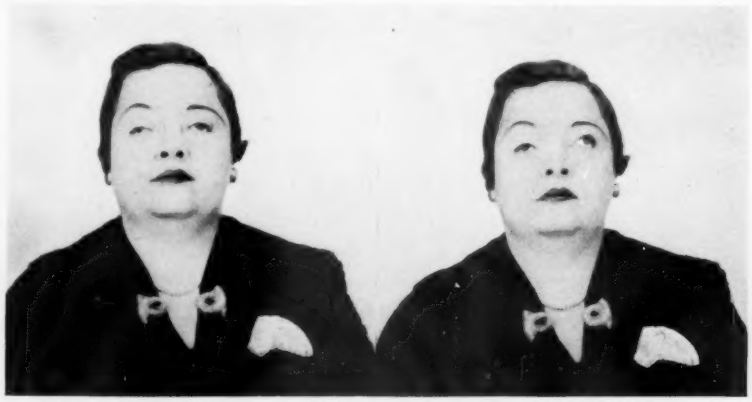

FIG. 11.—Photographs of patient (Case 3) taken eight months after the operation. It will be seen that there is slow but continued improvement in the ptosis and in the movements of the extra-ocular muscles supplied by the third nerve.

Intracranial aneurysms may be divided into two general groups: (1) Those within the substance of the brain; and (2) those on the surface of the base of the brain. The latter arise from the circle of Willis, or from the carotid in its intracranial course or within the carotid canal (and project into

the cranial chamber). The former are frequently larger, because a tissue wall withstands their progress; they are usually encountered during operations for brain tumors. The aneurysms at the base of the brain are usually relatively or actually small, are frequently multiple and their presence is known or suspected from five disturbances: (1) Recurring, sudden, severe pain and headache back of an eye; (2) subarachnoid hemorrhage; (3) palsies of the nerves to the extra-ocular muscles, particularly the third; (4) involvement of the trigeminal and sympathetic nerves; and (5) unilateral loss of vision with primary optic atrophy. One must always suspect an intracranial aneurysm when there are sudden, severe pains or headaches confined to one side and in the general region of the eye or temple. And the diagnosis becomes highly probable when one of the nerves entering the cavernous sinus is affected. The sudden severe pains or headaches will usually recur at relatively short intervals. The syndrome of recurring, sudden, severe pains in one eye, side of the head, or side of the face(frequently all are combined), plus ptosis and other evidence of disturbed function of the nerve, are almost pathognomonic of an aneurysm of the internal carotid artery or the posterior communicating artery. In each of the three aneurysms reported and in a fourth reported elsewhere, this syndrome was the sole basis for the diagnosis, and was regarded with sufficient certainty to warrant exploration of the aneurysm. This was also true in the case of Walsh and Love.[22] Although this syndrome has long been known and emphasized (Hutchinson,[9] Beadles,[3] Symonds,[19] Sands,[17] *etc.*), it has been only during the last few years that surgeons could feel sufficiently secure of its pathognomonic significance and at the same time of the surgical possibilities to permit exploration of the lesion.

Subarachnoid bleeding is with almost equal certainty an indication of an aneurysm on the circle of Willis, but rarely is there any indication of the location of the aneurysm, even to the right or left side of this vascular circle. The eyegrounds may give an indication. Not infrequently a huge round hemorrhage develops in the disk or retina in addition to many other smaller ones. This picture is pathognomonic of a subarachnoid hemorrhage, and in two recent cases the major hemorrhages were on the side of the aneurysm. A rapidly developing choked disk is usually also present and this, too, is more advanced on the side of the aneurysm. However, these indications are usually in the very severe hemorrhages, or are usually late manifestations. Ophthalmic migraine is often due to aneurysms of the carotid artery and should always be borne in mind as the probable cause.

Roentgenologic examination of the head is at times helpful in the differential diagnosis of intracranial aneurysms. Linear, curved shadows always suggest an aneurysm, and several examples of this have been demonstrated. Erosions of the landmarks of the sella and middle fossa of the skull, have resulted from large aneurysms of the carotid, usually arising within the cavernous sinus. Heuer and the writer[8] described such a case in 1916; Schüller another, in 1918; both were substantiated by necropsy. Sosman and Vogt (1926) added three more. Other writers contributing on this subject

are McKinney, Acree and Soltz[13] (1936), and Jefferson (1938). In one of Jefferson's cases the shadow appeared seven years after her first symptom and seven years before death.

Arteriography, introduced by Moniz, in 1937, gives every promise of the greatest help in precisely diagnosing and localizing intracranial aneurysms. Already, quite a number have been so graphically demonstrated by the intra-arterial (internal or common carotid) injections of thorotrast that one cannot be skeptical of its importance. A glance at the beautiful roentgenograms, with the sharply defined and unmistakable aneurysms, shown by Dott, Tönnis and Jefferson, after these injections, is absolutely convincing. So graphic are they, that nothing is left to the imagination. And yet one wonders whether the absence of an aneurysmal shadow can be accepted as positive evidence that an aneurysm does not exist, *i.e.*, whether one can exclude an aneurysm on negative evidence. Also, is a bilateral internal carotid injection indicated after negative single injections, and even then would bilateral negative injections be absolute evidence in excluding an aneurysm? I must confess a great reluctance to use arteriography, fearing thrombosis of the big arterial trunk, or some cerebral complication from thrombosis of one of the smaller trunks, possibly even the induction of hemorrhages from the aneurysm. This may well be a prejudice of one who withholds all accessory methods in diagnosis, even lumbar punctures and ventriculography, unless there are very necessary indications for their employment. It is always my feeling that the least done to a patient, the better. Complications are bound to arise from time to time as a result of seemingly trivial accessory examinations (lumbar punctures and air injections, intravenous injections, *etc.*, for example) and the patient may lose his life as a result. I realize full well that no bad results have been reported from the use of arteriography, but when deaths have occurred in patients who have previously had injections of thorotrast, one wonders whether this method can be excluded as a factor, or at least whether in the very ill state of the patients they would not have been better off without the additional insult, even though reportedly a harmless one. There can be no quarrels in its employment when the silent aneurysms causing subarachnoid hemorrhages are sought, but I can see little, if any, reason for its employment when the history and localizing neurologic evidence is so convincing. Even in the group associated with subarachnoid hemorrhages, without any indication of the side of the lesion, there is room for disagreement. So many of these patients recover from the immediate effects of the hemorrhage, and go for many years without further trouble, that one is reluctant to add to the immediate risk. I have seen many such cases. On the other hand, only recently, I advised against a vascular injection of thorotrast when the patient was recovering and was seemingly well after a subarachnoid injection. A week later she had another hemorrhage and death resulted promptly. Unquestionably, precision and elimination of guesswork will eventually be the better course with lesions so grave and at the same time so capricious in their course. And the attainment of this end can, from the present outlook, be

only through arteriography. Doubtless, with time, the writer will use it more, but only when there is no indication whatever of the side of the lesion. At present, however, if by headache, pains or other subjective sensation there is indication of the side of the circle on which the aneurysm is located, I should prefer (in cases with subarachnoid hemorrhage) operative exposure of the base of the brain on that side, rather than arterial injections. However, it is only fair to add that since the type of operation to be employed in the treatment of intracranial aneurysms differs with the surgeon, the reaction to the use of angiography may differ accordingly. Those who feel that ligation of the carotid in the neck is safer, and on the whole preferable to direct inspection of the aneurysm, may feel more justification in viewing the aneurysm roentgenologically.

Treatment of Arterial Aneurysms.—Since the treatment of arterial aneurysms is still in its infancy, one cannot be dogmatic concerning any method of handling them, or indeed, in many instances, that surgery should be undertaken. That a certain number of aneurysms have been cured spontaneously from thrombosis is perhaps true, although one cannot be too certain of this. The long absence of symptoms leads us to suspect a cure, but subsequently, much of the false aneurysmal sac is largely filled by a laminated clot when the mouth of the aneurysm and a small portion of the sac adjoining the mouth remain patent; and the aneurysm is not only actually still active but may be of tremendous size. Although further bleeding may not occur and the patient may be symptomless for a long period of time, the silence may well be only deceptive. I have seen several such examples within the brain substance and the aneurysm has gained considerable size—even to the proportion of a large tumor—gradually and doubtlessly paroxysmally, and yet without external bleeding or other symptoms. In this state of apparent rest, the aneurysm is merely bleeding into the false sac and behind a thrombotic wall which is slowly extending; under such circumstances the aneurysmal cavity may be very small. This is probably more true of aneurysms within the brain substance than those on the exterior. Such findings lead one to suspect that in many instances the reputed cure of the aneurysm may be more apparent than real. However this may be, one is reluctant to advise surgical treatment for aneurysms on the circle of Willis unless the subarachnoid hemorrhages are sufficiently severe to endanger life and are known to recur.

For aneurysms of the carotid producing oculomotor palsies and severe recurring pains there can be no doubt concerning the need for surgical intervention. Nor is there, in safe hands, such a formidable risk as the subject of aneurysms might suggest. However, for aneurysms elsewhere along the circle of Willis one must assume at the present time, in the absence of surgical tests, that the risk is greater and the possibilities of aneurysms favorable for treatment are undoubtedly less. Many of them involve both the carotid and anterior or posterior communicating or the middle cerebral arteries, making the eradication or the trapping of the aneurysm seemingly impossible. Then, too, these aneurysms must be localized, at least, to the side of the circle of Willis. In

most instances this determination can be arrived at only by arteriography. The risk attending surgical efforts must, therefore, depend upon the exact site and the size of the aneurysm. There is no reason to believe that the surgical treatment of aneurysms of the anterior cerebral artery, or even the anterior communicating artery, should be attended by any more risk than one of the carotid, and probably not so much, but aneurysms of the posterior communicating or posterior cerebral or basilar arteries would be much more difficult to expose sufficiently to "clip" the trunk of the artery on both sides of the aneurysm. However, if nothing more could be done, it is not impossible that clipping of the arterial trunk on one side of the aneurysm may be adequate.

For aneurysms of the internal carotid artery, either within the carotid canal or within the cranium, the best type of surgical treatment can only be evolved from experience. Unquestionably, the best treatment for such aneurysms within the cranial chamber is "clipping" the neck of the aneurysm with or without coagulation of the sac beyond the clip. The great advantages of this method are, of course, that the treatment is a direct one with an absolute assurance of cure, and that the internal carotid artery is not sacrificed. But from a pathologic study of quite a number of these aneurysms, it is hardly conceivable that many of them are so favorably situated and constructed that closure of the neck will be possible. It is not impossible that the application of a silver clip to the body of a small aneurysm, that exhibits no appreciable neck, may not suffice, or even the application of the cautery to the aneurysm, without an intermediary clip, may prove to cure an aneurysm that otherwise would appear hopeless. Opening and packing an aneurysm such as was done by Tervani and Cushing is far more hazardous and more difficult. Certainly this method of attack would be a last resort.

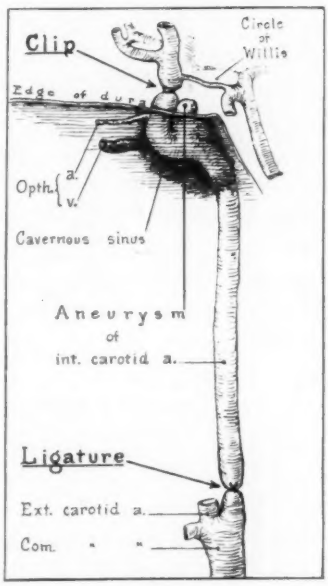

FIG. 12.—Diagrammatic representation indicating the position of the aneurysm and the method of surgical attack upon it, by trapping the aneurysm between the carotid, intracranially, and the ligature of the internal carotid artery in the neck.

For those aneurysms of this vessel that are within the cranial chamber and below the main branches, or are within the carotid canal, two lines of attack may be used: (1) Ligation of the internal carotid in the neck—Hunter's operation of ligation in continuity; and (2) ligation of the internal carotid, both within the cranial chamber and in the neck, *i.e.*, trapping the aneurysm between two ligatures. (Fig. 12.) It is assumed that the diagnosis can be established in these cases solely by the history and the neurologic examination. The three cases included in this report have been operated upon by the latter

method. Five of Dott's carotid aneurysms have been operated upon by the first method, three surviving and two ending fatally; however, since his deaths were in very ill patients, the ligation of the carotid may or may not have been contributory. However, both of his deaths were in patients whose ages, 49 and 50, were beyond the safe limit for total ligation of the internal carotid. My reasons for the intracranial approach first and ligation of the carotid at that site, have been that one could see the aneurysm and perhaps treat it by ligating the sac alone without sacrificing the internal carotid artery. And at least one can be certain of the character of the lesion, for tumors not infrequently must be considered in the differential diagnosis. Ligation of the neck of the sac has been possible in one of four cases. On the other hand, it may well be preferable to define the exact position and size of the aneurysm by arteriography and be guided in the subsequent method of surgical treatment by these results. Doubtless, to many, perhaps most surgeons, such a precise test will be preferable to a cranial exposure which, if ligation of the carotid in the neck should prove to be sufficient in producing a cure, may in the end be unnecessary. One's reaction to these alternative views will eventually depend upon: (1) Whether or not arteriography carries more or less danger than the cranial exposure; and (2) whether the degree of assurance of simple cervical ligation is sufficiently great.

The cure of an aneurysm by ligation of the internal carotid in the neck (in continuity) is thought to be dependent upon the development of a thrombus due to lowered blood pressure. The thrombus is presumed to be formed in the aneurysmal sac and either solely there or as a secondary extension of a thrombus extending from the site of the ligature in the internal carotid to and including the aneurysm. Our knowledge of thrombus formation in vessels with normal walls and without infection is none too secure. That such a progressive thrombus does form in the presence of infection or a ruptured intima is well recognized, but neither in experiments upon animals nor in clean human ligations, is there any certain evidence that a thrombus develops and spreads from the point of the ligature. Recently, I have seen at necropsy, two internal carotid arteries that have been ligated with silk, one three months, the other six weeks, and there was no gross thrombus formation in one and a tiny one, just visible to the eye, in the other. And in another case, I explored and opened an internal carotid artery in the neck just above a total ligature of this vessel, that had been placed several months earlier, and it not only bled freely but had no thrombus at that point. On the other hand, I have seen a thrombus develop spontaneously from a defective calcified wall in the cavernous portion of the internal carotid with a break in the intima and spread far down the neck and upward into the three intracranial branches of this vessel. In one of Dott's cases, a thrombus was said to have extended from the site of a cervical ligature on the internal carotid almost to the intracranial division of the carotid and including the aneurysm. However, since death resulted only 18 hours after the ligature was placed, one wonders

whether, after such a short interval, it is possible to differentiate with any degree of certainty between a postoperative and a postmortem thrombus.

In a recent publication, the writer[4] has shown that, in an analysis of many cases of carotid-cavernous arteriovenous aneurysms treated by ligation of the internal carotid artery in the neck, only about one-third are cured either immediately or within a short period thereafter. The cure of these aneurysms, when it occurs, is by the advent of thrombosis, just as in arterial aneurysms. But in every reported case of arterial aneurysm that has survived ligation of the internal carotid artery in the neck (three by Dott, one by Walsh and Love, and one by Jefferson) a seeming cure has resulted, and promptly. These results are far superior to those obtained by proximal ligation for aneurysms affecting other large arteries, where indeed the resultant cure is unusual. Certainly this difference in results cannot be due to a lesser collateral blood supply because the return of blood from the circle of Willis is very prompt and great. If these results hold even approximately, arterial aneurysms are decidedly easier to cure by ligation of the internal carotid than the arteriovenous type. And if subsequent tests prove this to be true intracranial ligation of the internal carotid artery would be unnecessary. At least, it can be withheld until the cervical ligation has proved to be ineffective. Possibly one should be prepared to believe that the cure of arterial aneurysms following this line of treatment would be more frequent than in the arteriovenous variety because they are blind-end sacs; whereas, in the latter there is only a thin-walled fistula in a continuous vascular channel, and through this the continuity of blood flow would be more probable. This alone will answer the question of the permanency of cures by simple ligation. For the moment, the cures appear to be no less perfect than by the double ligation.

Moreover, cervical ligations of the carotid can be effective only when the aneurysms are in certain locations. When situated in certain other positions, it may be very dangerous, *i.e.*, where the collateral circulation through the circle of Willis may be compromised or congenitally inadequate. In the case reported by Albright,[2] in 1929, I had tried this method of tying the internal carotid in the neck, using, however, the seemingly safer partial closure with a fascial band instead of a total occlusion with a ligature. This was done because the total occlusion test (by finger compression) indicated that complete closure of the internal carotid artery would not be tolerated. About eight hours after the operation (which was performed under local anesthesia), cerebral signs began to appear and steadily progressed. Death resulted three and one-half days after the operation, despite the removal of the fascial band within two hours after the first cerebral sign. There was no suggestion of a thrombus within the carotid artery or its branches. Cerebral softening resulted in the part of the brain supplied by the middle cerebral artery. Cerebral anemia was found at necropsy to be due to pressure of the aneurysm, which was a fairly large one, on both the middle cerebral and the posterior communicating arteries, thus precluding adequate collateral blood supply to the brain.

When an aneurysm is located along the circle of Willis and is so situated and is large enough to compromise or destroy by compression the circulation in the posterior communicating or middle cerebral or anterior communicating arteries, it is quite probable that the middle cerebral artery will receive an insufficient blood flow and the result will be cerebral softening. The above case is an example and is probably not exceptional. Dott came to a similar conclusion from his arteriographic studies. Then, too, one of these collateral trunks may be congenitally too small to play its part in maintaining the collateral circulation. It is doubtful how dependable the preoperative test of occluding the carotid artery with the finger may be, especially when it is negative, for in the above case the test was negative and it was eight hours before the effects of cerebral anemia were manifest. The importance of this test lies in detecting those cases in which an immediate effect is produced by closing the carotid.

Another reason for preferring exploration of the lesion, is that tumors at times may be disclosed when an aneurysm is suspected. Frequently the differential diagnosis between an aneurysm and tumor must be considered unless, of course, arteriography is used. And finally in reviewing the fatal case reported by Albright, I wonder if a direct attack upon the aneurysm might not now offer hope of success.

The age of a patient will always be an important determining factor in the line of treatment. After the age of 40 or 45 total ligation of the internal carotid artery unquestionably carries an increasing hazard from cerebral complications, due to inadequate blood supply. Before the age of 40 or 45 ligation of this important trunk is usually without adverse effect, though this is not always true, as our cerebral anemia from partial ligation demonstrates; this patient was only 33.

To protect patients over 40—indeed, all ages—the carotid compression is indispensable; it should be carried out for ten minutes without interruption. The importance of this test has been repeatedly emphasized by Matas.[15] It does not insure against eventual trouble, such as has been noted above, but when positive it does pick out individuals in whom ligation of the carotid would certainly be followed by hemiplegia and probably death. If, therefore, an aneurysm of the brain is suspected, the primary exposure of the aneurysm would not be attempted unless this test were negative. And, therefore, if the test were positive one would be forced to forego the intracranial exposure. But total ligation of the carotid in the neck would be equally contraindicated. The only safe surgical attack would then be, first a partial occlusion of the carotid in the neck, with a band of some type; I prefer a band of fascia lata. Four to six weeks later this artery can with safety be totally occluded. Finally, if there remains evidence of the patency of the aneurysm the intracranial exposure and attack can be carried out without risk of inadequate cerebral blood supply. That the age at which the carotid can be ligated with impunity varies with different individuals is shown by the fact that Magnus[14] ligated the internal carotid artery in a patient 69 years old, and McKendree and

Doshay[12] report another ligation by Elsberg in a patient age 58; Jefferson's case was 57. The end-result, of course, depends entirely upon the amount of collateral circulation through the anterior cerebral and posterior communicating arteries, and this will always be a variable factor regardless of age.

Results.—In Case 2, the return of all functions and the disappearance of all symptoms were prompt and complete. Within six weeks the eyelid and all of the movements of the extra-ocular muscles were perfectly normal. In Case 1, the symptomatic cure was just as prompt and complete but the return of the muscular function in the lids and eyeballs has been slow and two years after the operation is still incomplete (Fig. 3). In Case 3, all symptoms disappeared immediately. It was nearly three months before the eyelid began to elevate and at the end of four and one-half months the first return of movement in abduction of the eyeball had begun; other movements were as yet unchanged. The great difference in the rapidity with which the ptosis and extra-ocular palsies have cleared is noteworthy. The explanation can only be conjectured. It is noteworthy that the two cases in which the beginning and rate of improvement has been slower, had absolutely complete loss of all functions referable to the third nerve before operation; whereas, the patient whose oculomotor functions cleared so quickly and so rapidly had only weakness and not total paralysis of these functions. Our conclusion is that in the latter case the aneurysm had only pressed upon the nerve, whereas, in the other two, more slowly improving cases, the third nerve was probably incorporated in the wall of the aneurysm within the cavernous sinus.

The visual changes following the operations have also been interesting. In the paper on the treatment of carotid-cavernous arteriovenous aneurysms it was shown that the same two ligations of the internal carotid artery, *i.e.,* in the neck and intracranially, did not affect the vision on the affected side. Since the ophthalmic artery arises from the carotid between the two ligatures, the effect upon vision was anxiously awaited and fortunately proved to be unaffected. Doubtless because there was adequate collateral circulation from the branches of the external carotid within the orbit. From the three cases in this series, the vision remained normal in two; in fact it was greatly improved in one, *i.e.,* from 20/50 to 20/20. In the remaining case, Case 1, however, the vision was totally lost. In this case a plug of muscle was introduced into the carotid (Brooks' method) in the neck. It would seem reasonable to believe that from this a propagating thrombus had developed and incorporated the lumen of the ophthalmic artery and perhaps even of the retinal artery. Proof of this is, of course, not obtainable, but in none of the six other patients with the same double ligation was vision made worse.

SUMMARY AND CONCLUSIONS

(1) Three cases of arterial aneurysms of the intracranial portion of the internal carotid artery are presented. In each the aneurysm was alongside the carotid as it came through the cavernous sinus.

(2) Each was treated by ligating the internal carotid artery, both intra-

cranially (with a clip) and in the neck (with a ligature of silk). All are symptomatically well.

(3) The use of arteriography to disclose the aneurysm is discussed. It is a valuable diagnostic adjunct in silent aneurysms with subarachnoid bleeding. At the present time its use would appear to be indicated only when a diagnosis is impossible by other means. Perhaps its more frequent employment in precise localization and size of the aneurysm when its diagnosis is clear may be in the offing.

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DISCUSSION.—DR. ERNEST SACHS (St. Louis, Mo.): I would like to recall three cases in which I put a band on the internal carotid to control aneurysm. Two did well; the third one is of particular significance because it emphasizes the point Doctor Dandy made, that even if you try a preliminary compression for 15 minutes, it may not be an indication that occlusion of the artery will not lead to trouble. This particular case was treated for some time, compressing the artery, and there was no discomfort; so we put on a Matas band, and she was comfortable for 36 hours. Then suddenly, in the middle of the night, she developed a hemiplegia and, in spite of the fact that we removed the band the hemiplegia persisted. She died a few days later.

I have exposed several aneurysms intracranially, but they have been so large that it was impossible to do anything with them. I think it is usually worth considering getting a result by ligating the internal carotid, as it is a simple procedure and in a certain number of cases will relieve the patient.

DR. E. F. FINCHER (Atlanta, Ga.): I had the privilege some several months ago of hearing Doctor Dandy tell of his success in treating, intracranially, his first case of a carotid aneurysm by placing a silver clip on the neck of the aneurysmal sac. At that time, he anticipated that such a procedure was not always possible and suggested, should the problem demand it, that an intracranial aneurysm might be "trapped" by ligation of the internal carotid artery in the neck and then applying a silver clip to the intracranial carotid vessel, about the aneurysm. I did not know until I listened to his paper to-day that he had carried out such a procedure, so I have nothing to add other than another case of an intracranial aneurysm of the carotid artery successfully treated by operation.

Briefly, the history is that of a male, age 32, who entered Emory University Hospital, having had three vascular "blow-outs." The fourth aneurysmal leakage occurred on the morning of the day in which his thorotrast injection had been scheduled. This fourth "accident" resulted in a hemiplegia. The hemiplegia had practically disappeared by the tenth day, and the thorotrast injection was carried out under direct visualization, which demonstrated very plainly the aneurysm, which had been previously diagnosed clinically from the history and neurologic examination. Immediately following the roentgenologic examination, the internal carotid vessel was ligated in the neck. After a lapse of two weeks a craniotomy was performed, exposing the carotid artery just beneath the optic nerve. A silver clip was placed on the vessel anterior to the anterior communicating artery, and a second clip anterior to this one where a break in the vessel was seen. The bone flap was a more extensive one than would be necessary for adequate intracranial exposure of the carotid artery, but was employed because of the fact that the patient had just recovered from a hemiplegia, and, also, because of a fear of too excessive traction on the frontal lobe, which was to be avoided.

This patient was discharged from the hospital three weeks after the craniotomy, at which time he was beginning to voluntarily elevate the para-

lyzed eyelid. I was very glad to hear Doctor Dandy tell of the lapse of time from operation until recovery of ocular activities, for I now feel that the patient here reported has a definite chance for complete recovery of his ophthalmoplegia.

DR. I. M. GAGE (New Orleans, La.): I wish to congratulate Doctor Dandy on his most excellent presentation dealing with intracranial arterial aneurysms. I believe, with him, that the incidence of arterial aneurysms of the internal carotid artery, occurring intracranially, will increase when the signs and symptoms of this condition are more thoroughly understood by the profession. I believe that a number of cases of trigeminal neuralgia are due to arterial aneurysms of the internal carotid artery which are not recognized. The cases presented by Doctor Dandy at a previous meeting of this Society and the ones presented to-day demonstrate conclusively that a clinical diagnosis is not so difficult provided the symptomatology is kept in mind. He has also given to us a method of treatment which is superior to all other methods that have been used, *i.e.*, "trapping" the aneurysm between two ligatures.

Doctor Dandy stated that the successful outcome of the treatment depends upon the collateral circulation. If the collateral circulation is inadequate, there always occur varying degrees of cerebral anemia with partial or complete hemiplegia. The mortality is proportionate to the presence or absence of an adequate collateral circulation. If the collateral circulation is inadequate, the mortality following ligation of the internal carotid becomes almost prohibitive.

Therefore, before any attempt to ligate the internal carotid artery for intracranial aneurysm of the internal carotid artery, the presence or absence of a definite and adequate collateral circulation must be determined. This can be definitely elicited by the Matas test. This test has been demonstrated and explained by Doctor Matas before this Society many times. It consists of applying the Matas compressor to the common carotid artery, obliterating the same, and noting the occurrence of cerebral manifestations. If the patient can withstand obstruction of the common carotid artery for half an hour or more, it is safe to proceed with the ligation of the internal carotid. If the patient cannot tolerate the compressor, and exhibits cerebral symptoms, the collateral circulation is inadequate. This inadequacy can be overcome by developing the collateral circulation by applying the Matas compressor daily for increasing lengths of time.

In 1934, before this Society, I advocated blocking the sympathetic ganglia as an aid in developing the collateral circulation in all cases of pure arterial aneurysms of the peripheral arteries. The success I have had with this method in eight cases of arterial aneurysm of the peripheral arteries has surpassed my expectation, and the results have been most gratifying.

Therefore, in developing the collateral circulation before ligating the internal carotid artery, I would advocate that the stellate ganglion on the same side be injected with novocain. This will relieve the vasomotor spasm which is concomitantly present in diseases of the artery. It will be necessary to repeat the injection of the ganglion if novocain alone is used. However, if one uses pontocaine in almond oil, the effect lasts for several days. This injection should be done both preoperatively and immediately postoperatively, and every three to four days following the ligation.

The effect of stellate ganglion injection on the collateral circulation can be estimated preoperatively by applying the Matas compressor and noting the cerebral reaction. The compressor is then removed and the stellate ganglion is injected with 1 per cent pontocaine. After one-half to three-quarters of an

hour, the compressor is reapplied and the reaction of the cerebral circulation is noted. If the response is marked, *i.e.*, no cerebral manifestations, then one may be justified in certain cases to section the preganglionic fibers of the stellate ganglion surgically.

Doctor Dandy's operation of "trapping" the aneurysm between two ligatures has resulted in permanent cures in all the cases in which the procedure has been employed. If the collateral circulation is adequate, cerebral complications resulting from deficient blood supply will be greatly minimized.

DR. GILBERT HORRAX (Boston, Mass.): One thing about aneurysms has not been mentioned—the chances of recurrence. It is difficult to know when to explore these patients, since many of them recover fully—even from a third nerve paralysis—and may never have a similar episode. In one or two cases, we have done what Jefferson has done, namely, pack small pieces of muscle all around the aneurysm to prevent leakage, where ligation is not feasible.

THYROTOXICOSIS

INCLUDING A STUDY OF THE DURATION OF PREOPERATIVE TREATMENT

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SINCE the introduction of iodine administration in the preparation for thyroidectomy of patients with thyrotoxicosis, the general principle governing the duration of preoperative treatment has been the continuance of such treatment until the patient's condition has improved to a degree somewhat vaguely expressed as "optimal." Theoretically, the patient is in this state when his symptoms and signs can be no further improved by continued treatment, his pulse rate and his basal metabolic rate can be no further lowered and his weight is stationary or increasing. It is the generally accepted belief that, after the optimum point is reached, further delay in operation is harmful. One school of thought attributes the harm directly to iodine medication, maintaining that an oversupply will stimulate the gland to renewed hyperactivity. Another school attributes the harm not to the iodine itself; it believes that, near the optimum point, the low level of a cyclic variation in the intensity of the disease has been reached, and that, although the iodine still has some effect, it cannot control the progress of a rising wave of thyrotoxicosis. Under either explanation, it is clinically important to determine as accurately as possible when the optimum point has been reached.

The obvious difficulty in this determination resides in the fact that, except in the occasional case in which all thyrotoxic manifestations disappear under treatment, the optimum point cannot be definitely known until it is past, when increasing signs of hyperthyroidism may appear. This difficulty is reflected in the literature, where only indefinite criteria for the optimum point are described. It would be useful if more accurate quantitative means were available of measuring in advance the optimum duration of treatment, than the clinical impression that the individual patient has done as well as he will do.

On the basis of scattered clinical experiences, it had seemed possible to us that the violence and duration of the postoperative reaction might be inversely related to the duration of preoperative treatment; in other words, that the longer the period of preoperative treatment within reasonable limits, the less violent the postoperative manifestations of thyrotoxicosis and the shorter the duration of the postoperative reaction. If such a relationship could be demonstrated, the indications governing the length of preoperative treatment would be clarified, although, of course, the optimum point in the individual case could be no more easily identified.

No previous reports have been found in which this comparison has been studied. In spite of the fact that the expected correlation was not demonstrated, certain conclusions from the present study are justified. The presentation will be divided into two independent portions: First, a study of the toxic thyroid problem in the University of Virginia Hospital, to serve as a background for the second, which is a detailed analysis of the relationship between variations in the duration of preoperative treatment and the patient's course.

Part I. Material and Mortality

This report is based on 401 cases of goiter treated by operation at the University Hospital, during the years 1925 to 1937 inclusive. During this period there were a total of 87,661 admissions to the hospital on all services; the admission rate was, therefore, approximately one case in 218. Of the 401 cases, 290 were classified as thyrotoxic. This is the group with which the present study concerns itself. A study of cancer of the thyroid during the same period is in preparation.[1]

Sex and Race.—There were 184 white and 47 colored females, 53 white and six colored males. There were, therefore, in all, 231 females and 59 males, 237 whites and 53 Negroes (Table I). The cases of thyrotoxicosis among Negroes represent 18.2 per cent of the total, which may be compared to the average hospital admission rate of Negroes with all diseases, namely, 16.6 per cent.

Table I

Analysis of Race and Sex Distribution in Relation to Mortality

	Number of Patients	Deaths	Percentage Mortality
Toxic			
White	237	2	0.84
Colored	53	5	9.43
Female	231	7	3.07
Male	59	0	0.0
White female	184	2	1.09
White male	53	0	0.0
Colored female	47	5	10.63
Colored male	6	0	0.0
Totals	290	7	2.41
Nontoxic	111	1	0.90
Totals	401	8	1.99

Age.—The distribution of the material by age is shown in Chart 1. The usual age difference between the diffuse and nodular groups is indicated. The greatest number of cases of the former occurred between the ages of 31 and 40, whereas the peak of the curve of incidence of the latter lies between 41 and 50 years.

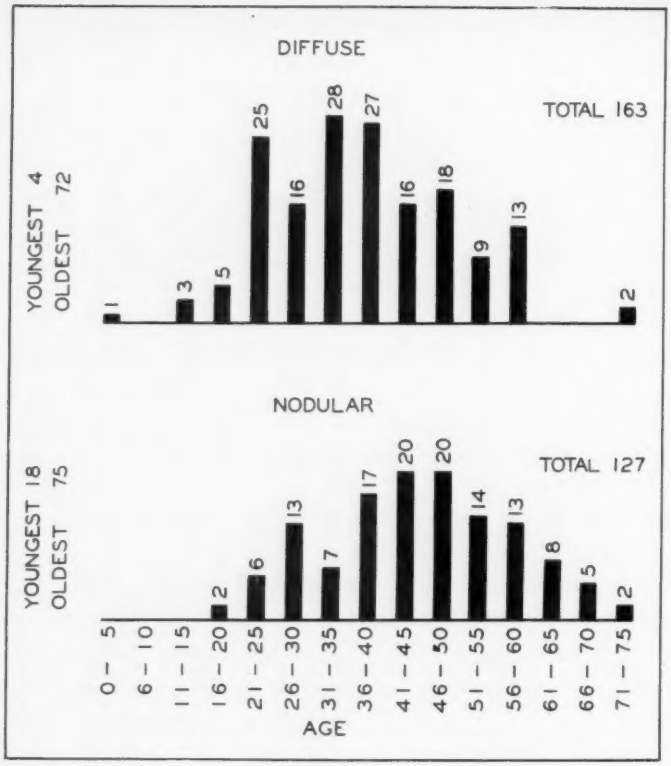

CHART 1.—Age distribution in 290 cases of thyrotoxicosis divided into diffuse and nodular groups.

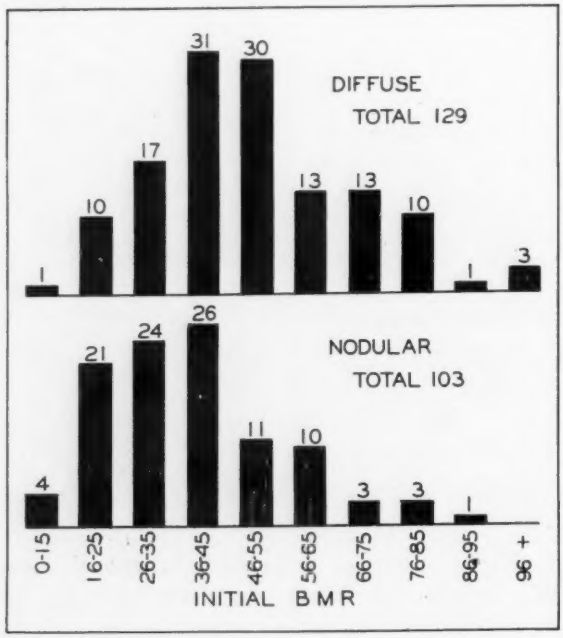

CHART 2.—Distribution according to initial basal metabolic rate readings of 232 cases of thyrotoxicosis, the iodine treatment of which began under hospital observation.

Degree of Toxicity.—On account of possible geographic differences in the problem of toxic goiter, an attempt to indicate the average severity of the disease in any series reported is pertinent. Although in the individual case, the level of the basal metabolic rate may be deceptive as an index of the severity of the patient's disease, yet this level is the only index that can be profitably presented in a quantitative form in the study of multiple cases. Chart 2, therefore, presenting the material divided by groups at various initial basal metabolic rate levels, will serve as a rough guide to the average severity of the disease as encountered at the University of Virginia Hospital. The entire series of cases is not represented in this chart, inasmuch as 47 (16.2 per cent) were admitted after iodine treatment by the family physician, and 11 had technically unsatisfactory first basal metabolic rate determinations. It will be noted that the peaks of the curves for both diffuse and nodular goiter occur at basal metabolic rates between plus 36 and plus 45. It will also be noted that there were treated 34 cases with metabolic rates above plus 65, an incidence of 14.6 per cent of the patients whose treatment began in the hospital.

Treatment.—Except for the cases requiring special consideration, such as thyrocardiacs and those admitted after iodine treatment elsewhere, the preoperative treatment was uniform. Rest in bed, a high carbohydrate diet, mild sedation and Lugol's solution comprise the general principles. The dosage of iodine was commonly 15 drops of the solution thrice daily, although some cases received a smaller amount. Many ward patients could not be protected from noise and excitement. The time for operation was chosen by consideration of six factors, namely: (1) Subjective improvement; (2) improvement in objective signs such as tremor, exophthalmos, motor activity, emotionalism, *etc.;* (3) the pulse curve; (4) the basal metabolic rate curve; (5) the weight curve; and (6) the changes in the consistency of the gland. The preferred operation was partial or subtotal thyroidectomy in one stage, the procedure becoming progressively more radical during the course of the period covered by the study. Only nine patients (3.1 per cent) had graded operative treatment. The medical and surgical services worked closely together in the study and care of the majority of patients.

Recurrences.—Eleven known recurrences, or 3.79 per cent, occurred. Of these, seven followed the treatment of 237 white patients (2.95 per cent) and four that of 53 Negroes (7.54 per cent).

Mortality.—The total patient mortality for the series is 2.41 per cent (Table I), representing seven deaths in 290 cases. Divided by race, the figures present a striking contrast. The mortality for the white patients is 0.84 per cent as contrasted with 9.43 per cent for the colored patients. It will be noted that among the 59 male patients there were no deaths.

Causes of Death.—Among the seven deaths (Table II), there occurred only a single death in thyroid crisis and a single death in congestive failure. One patient died of late thrombotic pulmonary embolism, and two deaths suggested air embolism. One patient died of asphyxia after return to the ward. Laryngeal paralysis or tracheal collapse was suspected. No tracheotomy was per-

TABLE II

ANALYSIS OF CAUSES OF DEATH

Hosp. No.	Year	Race	Sex	Age	Initial B.M.R.	Preop. B.M.R.	Interval after Operation	Apparent Cause of Death	Remarks	Autopsy
63351	1927	C.	F.	33	plus 78	plus 56	14 da.	Pulmonary embolism	Continuously febrile after operation	No
66728	1928	C.	F.	39	plus 66	plus 56	1 da.	Thyroid crisis	Had prolonged iodine treatment before admission	No
76082	1929	C.	F.	21	plus 16	plus 8	None	Embolism?	Marked blood loss at operation. Sudden death on return to ward	No
90103	1931	C.	F.	46	plus 52	plus 13	None	Air embolism	Sudden death after wounding vein	No
112452	1934	C.	F.	4	plus 52	plus 54	5 hrs.	Unknown	Enormous thymus found at autopsy	Yes
120986	1936	W.	F.	46	plus 33	plus 38	2 hrs.	Asphyxia	Sudden death. Laryngeal paralysis suspected	No
54531	1937	W.	F.	75	plus 22	plus 7	1 da.	Congestive failure	Cardiac of long standing. Risk deliberately taken	No

formed. The seventh case was a Negro child of four years, with an unexplained death several hours after operation. At autopsy an enormous thymus gland was found. It may be pointed out that in four of the seven fatal cases the level of the basal metabolism failed to drop satisfactorily before operation. In only one of these was the death apparently related to this phenomenon.

Comment.—The University of Virginia lies within 40 miles of a minor center of endemic goiter, the Shenandoah Valley. It is a general hospital, making no particular effort to attract thyroid disease. The hospital incidence of one in 218 may be contrasted with the hospital incidence at another Southern surgical center, the Charity Hospital at New Orleans, situated in an area where endemic goiter is not found. There, over a period of nine and one-half years, as reported by Maes, Boyce and McFetridge,[2] the incidence was one in 688, less than one-third of that reported herein. The incidence of thyroid disease among Negroes almost exactly equals the hospital admission rate of Negroes, a fact which supports the opinion that the black race is at least equally susceptible to thyroid diseases as the white.[2, 3, 4]

The opinion has been expressed by Maes and his colleagues[2] that in the Negro, particularly the male Negro, thyroid disease "presents a serious problem" and is "highly fatal," a conclusion based on their admittedly shockingly high mortality rates for this group. It was disappointing to find that the total mortality of the present series was 2.41 per cent, but further analysis of the data was reassuring, particularly in relation to the Charity Hospital findings. The mortality for whites was only 0.84 per cent, a figure that compares favorably with most modern mortality figures from the large northern clinics where the material is almost exclusively white (Table III). The Negro mortality of 9.43 per cent is, however, deplorable. In view of the experience of Maes and his coworkers, one is inclined to search for some racial cause for the difference. Certainly in the present series there have been no marked differences in the method of preparation for operation, the ward arrangements, the experience of the operators or the detailed observation and care of patients between the white and the black groups, at least insofar as the ward material is concerned. Nor do the causes of death offer any explanation. The only death in thyroid crisis, it is true, was in a Negro woman, but three other Negro deaths occurred, apparently not directly related to the degree of toxicity. Corroborative evidence that the disease may be surgically a separate problem in the colored race is furnished by the fact that the known recurrence rate among the Negroes was over twice as great as the known recurrence rate among the whites. One striking discrepancy with the Charity Hospital statistics may be noted. Whereas the highest mortality rate of all groups was there found among the Negro males, in the present series no deaths occurred among the colored men. As only six male Negroes were treated, this discrepancy is probably the result of chance alone. In this general connection the excellent record of Drennen[11] must be cited. He reports one death in a total of 203 cases, including 22 Negroes, none of whom died. Horsley's smaller series,[12] presenting no mortality, included no Negroes.

TABLE III

REPRESENTATIVE PATIENT MORTALITY STATISTICS

Author	Date	Locality	Number of Cases	Percentage Mortality
		Northern Clinics		
Moore[5]	1934	Seattle	1,860	0.86*
Pemberton[6]	1936	Rochester, Minn.	9,223 (exophthalmic only)	0.82
Lahey[7]	1935	Boston	6,345	1.10
Thompson, *et al.*[8]	1938	Chicago	181	1.65
Graham and Wallace[9]	1934	Edinburgh	125	3.20
Dunhill[10]	1937	London	?	2.60
		Southern Clinics		
Drennen[11]	1937	Birmingham	203	0.49*
Horsley[12]	1935	Richmond	118 (white)	0.00
Clifton[13]	1935	Atlanta	550	3.63
Maes, *et al.*[2]	1937	New Orleans	121	6.60
Davison and Poer[14]	1936	Atlanta	322	3.10*
Lehman and Shearburn	1938	Charlottesville	290	2.41
			237 (white)	0.84

* In none of the reports from northern clinics is the proportion of colored patients given; it is presumed to be minimal. The percentages marked with an asterisk have been calculated by the present authors from data given in the respective reports.

The question may be raised whether the present total mortality of 2.41 per cent could have been improved by increasing the low incidence of graded procedures (3.1 per cent). Again reference to the causes of death does not suggest that this is a major possibility. It cannot be denied, however, that at least one death might have been averted by a graded operative attack.

PART II. DURATION OF PREOPERATIVE TREATMENT

Scatter charts have been employed in studying the relationship of the various factors investigated. The cases represented on these charts form a selected group of 175 from which extraneous factors have been eliminated, such as treatment with iodine before entering the hospital, preceding or concurrent acute infections, iodism and voluntary delay in consenting to operation.

The severity of the postoperative reaction in the 175 cases was first studied in relation to the duration of preoperative iodine treatment (Chart 3). The chart indicates that the level of neither the highest postoperative pulse nor the highest postoperative temperature bears any relationship to the period of preoperative preparation. Some of the most violent reactions followed the longest preparation, and vice versa. No tendency toward grouping along a curve is manifest.

In view of the possibility that the varying severity of the disease in the gross material might prevent the detection of such a tendency in Chart 3, the same and other studies were made in a group of cases of approximately equally severe disease. Using the basal metabolic rate as a rough index of severity,

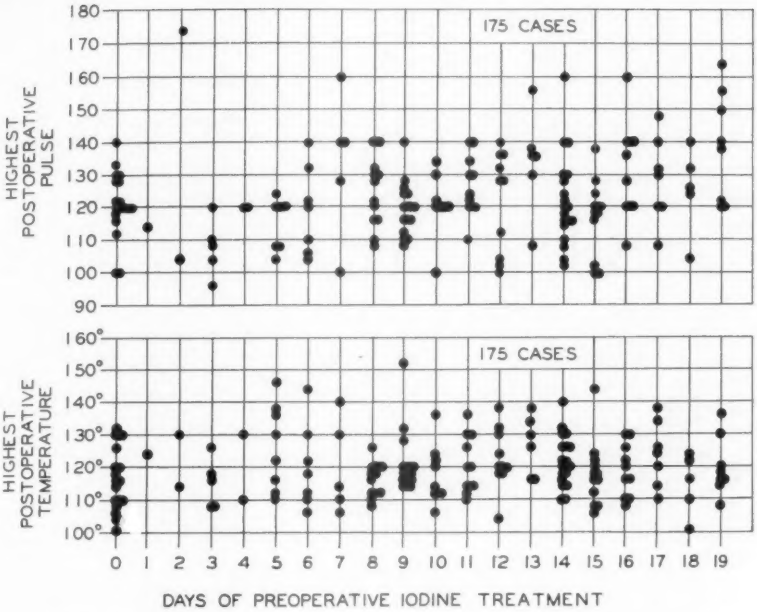

CHART 3.—Relationship of the highest postoperative pulse and temperature to the duration of preoperative iodine treatment in a group of 175 cases of thyrotoxicosis. See text for method of selection.

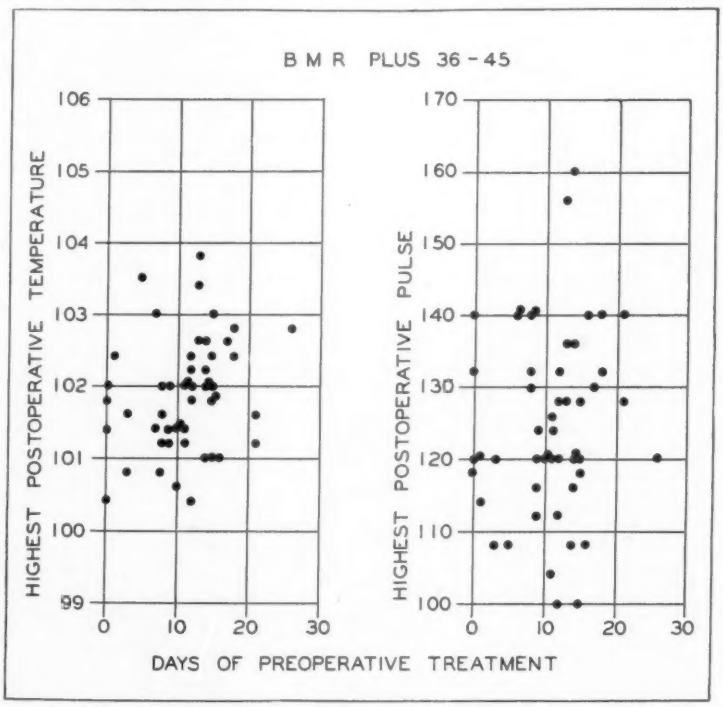

CHART 4.—Relationship of the highest postoperative temperature and pulse to the duration of preoperative iodine treatment in a group of cases of relatively uniform initial toxicity (initial B.M.R. plus 36 to plus 45). Cases were excluded as in Chart 3.

the largest group was chosen, namely, that with basal metabolic rates between plus 36 and plus 45 (Charts 4, 5 and 6). Again no correlation is seen. Length of preoperative treatment does not appear to be a factor in the severity of the

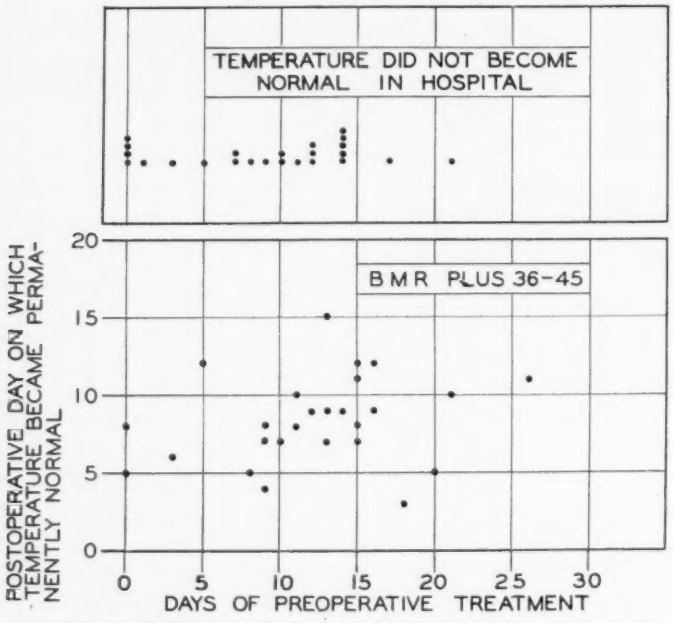

CHART 5.—Relationship of the duration of postoperative fever to the duration of preoperative iodine treatment in the same group of cases as that studied in Chart 4.

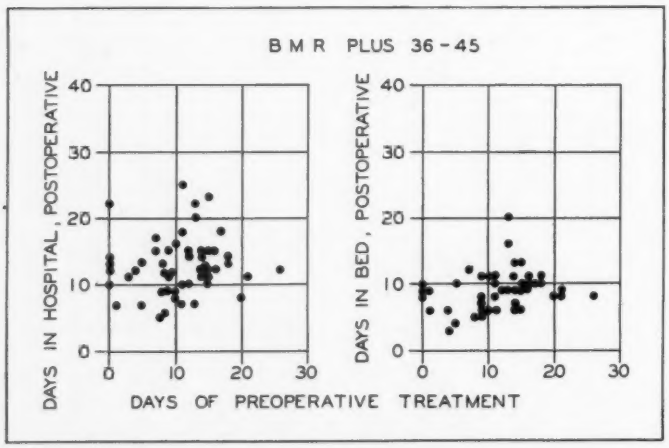

CHART 6.—Relationship of the duration of postoperative bed rest and postoperative confinement to the hospital to the duration of preoperative iodine treatment in the same group of cases as that studied in Chart 4. In several instances the charts did not indicate the first day on which the patient was allowed out of bed.

postoperative reaction as measured by the highest postoperative temperature and pulse (Chart 4), nor in the duration of the postoperative reaction, as

measured by the duration of fever (Chart 5) and the length of postoperative confinement in bed and in the hospital (Chart 6).

On considering this group of negative findings, it appeared pertinent to determine whether the initial severity of the disease might govern the violence of the postoperative reaction. This relationship was, therefore, investigated by plotting the initial basal metabolic rate against the highest postoperative temperature and pulse (Chart 7). Again it is obvious that there is no significant correlation either in the diffuse or in the nodular group. Many apparently mildly toxic cases suffered stormy postoperative courses, whereas

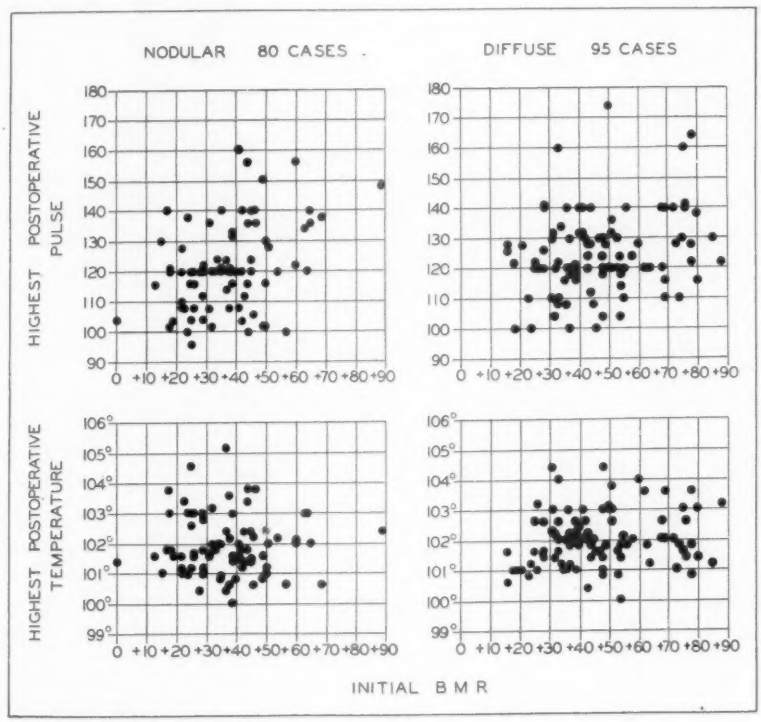

CHART 7.—Relationship of the highest postoperative pulse and temperature to the initial basal metabolic rate in the selected series of 175 cases, divided into nodular and diffuse groups.

the operations upon a number of cases with high initial basal metabolism readings were followed by minimal elevations of temperature and pulse.

It had been supposed that the initial basal metabolic reading must, in practice, bear a direct relationship to the duration of preoperative treatment. This conception was, therefore, finally investigated in the hope that it might illuminate the previous negative results (Chart 8). It is interesting to observe that no correlation whatever can be detected, either in the entire selected group of 175 cases or in the nodular and diffuse groups taken separately. Patients with initial basal metabolic readings just above the upper limits of the normal range were treated well into the third week, while apparently much more toxic

patients received treatment for eight days or less. The average duration of preoperative treatment in this series was just over ten days.

COMMENT.—From the failure to find any correlation whatever between the various factors studied, two or three deductions seem justifiable. In the first place the group of studies of the postoperative reaction following varying durations of preoperative iodine treatment indicate that there is no particular virtue inherent in prolonging the latter. One is not justified in assuming that an additional week of preoperative preparation after an operable state is reached will protect the patient from a severe postoperative reaction or save him a corresponding period of hospitalization during convalescence.

In the second place, the fact that the degree of postoperative storm is not related to the initial basal metabolic reading permits two possible interpreta-

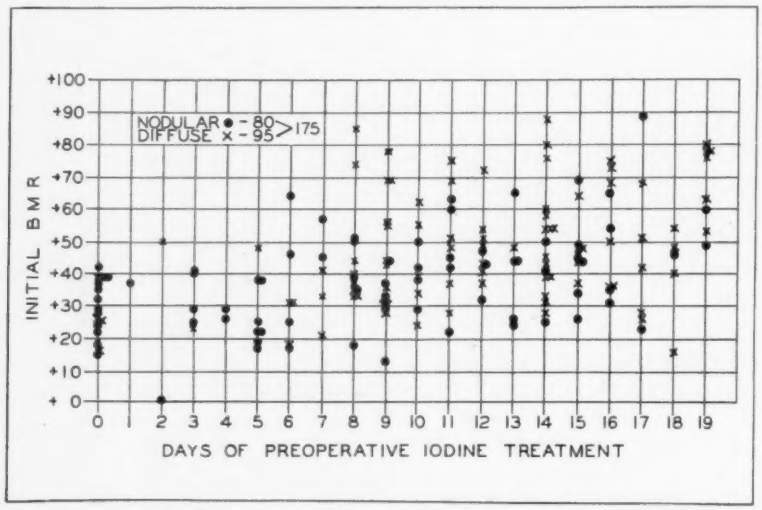

CHART 8.—Relationship of the initial basal metabolic rate to the duration of preoperative iodine treatment in the selected group of 175 cases. The nodular and diffuse cases are indicated separately by the designated symbols.

tions, each of which may play a part in the truth. One of these is the well-recognized fact that the basal metabolic rate by itself does not measure the illness of the patient or the surgical risk. The second interpretation is the probability that wide variations in the response of patients to preoperative treatment shuffle the cards so that many of the originally sickest patients come to operation as better risks than others less toxic at the beginning of treatment. It is probable that the coincidence of the period of preparation with a cyclic change in toxicity, downward in the cases that respond well and upward in those that respond more slowly, helps to explain the absence of correlation between the severity of the postoperative reaction and the initial basal metabolic level. It is frequently difficult to determine clinically in any case whether the patient is in a wave of increasing or decreasing toxicity; and totally impossible to express such determinations in the study of a series of cases.

Finally, all these negative observations lead to the conclusion that no quan-

titative rules for the handling of toxic thyroid disease can be laid down. That no such rules have been laid down in practice is indicated by the lack of correlation between the apparent degree of initial illness of the patient and the duration of his preparation for operation (Chart 8). Each case is a separate, individual problem. No criteria for operability can be expressed in any quantitative form and the optimum duration of preoperative preparation cannot be predicted. Authors are entirely justified in expressing only in the most general terms their opinions in regard to these two factors. The present data offer no promise that clear recognition of the optimal moment will ever become possible.

SUMMARY

(1) A series of 290 cases of thyrotoxicosis subjected to surgical operation is analyzed in the usual manner. Among 237 white patients the mortality was 0.84 per cent; among 53 Negroes, 9.43 per cent. The total mortality was 2.41 per cent. Known recurrence was over twice as great among the Negroes as among the whites. Graded operations were performed in 3.1 per cent of patients.

(2) A selected group from the same series was also analyzed by scatter diagrams in an attempt to study the possible relationship of: (a) Variations in the duration of preoperative iodine treatment and the patient's postoperative course; (b) the level of the initial basal metabolic rate and the patient's postoperative course; and (c) the level of the initial basal metabolic rate and the duration of preoperative iodine treatment. No correlation was found in any of the factors compared.

CONCLUSIONS

(1) That the surgical problem of thyrotoxicosis in the Negro probably differs from that in the white is indicated by a disproportionately high level of both the operative risk and the recurrence rate in the former.

(2) The severity of the postoperative reaction in the thyrotoxic is not lessened nor is the postoperative convalescence of the patient shortened by prolonging preoperative preparation beyond the point at which the patient becomes a good operative risk.

(3) No quantitative rules governing the duration of preoperative treatment can be laid down. Individualization is an essential element in the successful handling of toxic thyroid disease.

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DISCUSSION.—DR. FRANK H. LAHEY (Boston, Mass.): I would like to present some of our further experiences with blood iodine, because I think since I have spoken before on it, there have been some new observations which are of clinical value. Since we know that 65 per cent of thyrotoxin is iodine, it is reasonable to assume that if we have a resting thyroid we have

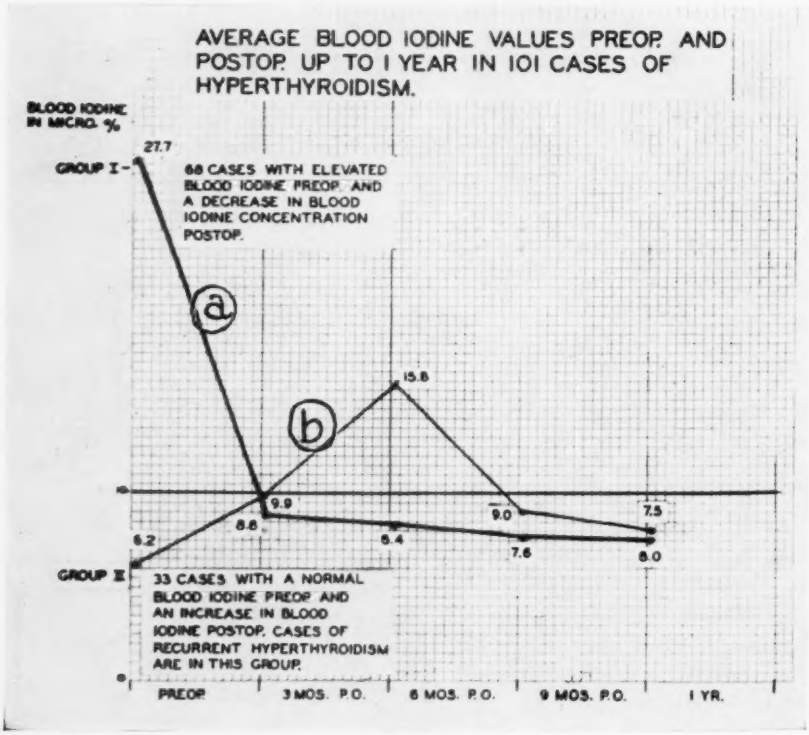

CHART 1.

a high content of iodine in the thyroid and a normal blood iodine. If we have an exophthalmic goiter, we should have a low content in the gland and a high blood iodine, and this is true in 70 per cent of cases.

We have superimposed the blood iodine upon the basal metabolism in 101 cases of exophthalmic goiter (Chart 1 *a*); the average basal metabolic rate in this group being +45 per cent, and the average blood iodine (normal, 10 micrograms per cent) being 22.8 micrograms per cent. At the end of six months after operation, both blood iodine and basal metabolism had become normal, descending in parallel lines. That is as it should be. We have been interested from a clinical point of view in determining in what group of cases recurrence

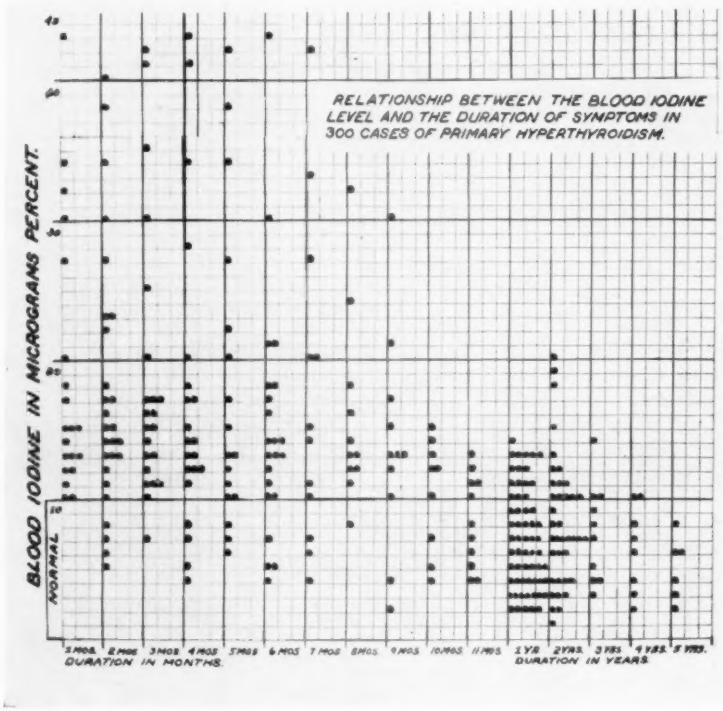

CHART 2.

of hyperthyroidism occurs, and in this 70 per cent group of typical high metabolism and typical high blood iodine there has been a percentage of recurrent hyperthyroidism in only 0.5 per cent. Furthermore, the number of patients in this group requiring multiple-stage operations is but 17 per cent.

In another group of patients with hyperthyroidism with a similar elevation of metabolism representing 30 per cent of all the cases, there is found a blood iodine which is normal or below normal, which by the sixth and ninth month after operation is slightly above normal and in a little over nine months after operation reaches normal (Chart 1 *b*). In this group the basal metabolism comes to normal typically as it does in the 70 per cent group. It is in this group that the high percentage of recurrence occurs, 22 per cent of these cases showing a recurrent hyperthyroidism. In this group multiple-stage operations are necessary; 45 per cent require multiple stages.

Here, then, are two laboratory measures from which can be obtained valuable clinical information. In the typical group with high metabolic rates and high blood iodine values, conservative subtotal thyroidectomy can be performed with a low percentage of multiple-stage operations; while in the atypical 30 per cent group having a high metabolic rate and low blood iodine value, radical subtotal thyroidectomy must be performed and a high percentage of multiple-stage operations.

Chart 2 is a scatter chart in which is related the height of the blood iodine to the length of time the patients have had the disease. These charts have been prepared by Mr. H. J. Perkin, a biochemist, who has been in charge of the iodine investigation in the Research Department of the Lahey Clinic.

It is to be noted that up to the time patients have had hyperthyroidism one year, most of the blood iodines are above normal, but that from one year on, most of the blood iodines are normal or below normal. The explanation of this is that the intake of iodine is constant, so that with a high blood iodine there is a high urinary output, establishing thus a negative iodine balance resulting in exhaustion of the iodine reserve.

Dr. Willard Bartlett, Jr. (St. Louis, Mo.): The aspect of the problem of thyrotoxicosis which Doctor Lehman has presented is so close to my heart that I cannot refrain from commenting on it, though from a somewhat different point of view:

Primary Criteria	Contraindications
1. Circulation	Decompensation
2. Nutrition	Rapid, continuing loss
3. Nervous	Acute psychosis
4. Excretion	Vomiting, diarrhea, sweating
5. Metabolism	Rising
6. D. V. A.	—

These criteria list organ systems and methods of examination through which extremes of the disease are primarily manifested and, in certain instances, become objectively measurable. Such extreme manifestations of *acuity* constitute absolute contraindications to operation during their continuance. This statement is made with complete disregard of type and duration of previous treatment; such considerations are secondary if we are to individualize these patients.

The last criterion refers to "duration of voluntary apnea," our breath-holding test which, more than any other single test, lets us estimate operative risk in mathematical terms. The complete plan of estimation of risk was established from a study of our mortality in thyrotoxic patients for the period 1926–1930; during those years the preoperative deaths formed 40 per cent of the total mortality, the postoperative, 60 per cent. In the five-year period subsequent to the establishment of the plan, the preoperative portion of total mortality rose to 65 per cent and the postoperative portion fell to 35 per cent. Better judgment was obviously being shown. Incidentally, the operative mortality was cut in half. But beyond this, the point is that if such a plan is used, operative deaths are predictable in all cases, and such has been true in our own experience where, with all the data carefully gathered before us, we have still violated one to three of our own criteria in an occasional case and a fatality has ensued. This is not a confession of ignorance, but of failure to "use what we know."

Too many medical students are still being taught that the maximum effect

of iodine is obtained in ten days and that thyroidectomy should be performed at such an interval after starting treatment. It is not quite as simple as that. I hope that Doctor Lehman's conclusion regarding the impossibility of generalizing as to the duration of preoperative treatment will gain the wide circulation it deserves.

DR. I. A. BIGGER (Richmond, Va.): The only point I wish to comment upon is Doctor Lehman's statistics regarding the mortality rate in the colored and white groups. We have had the same experience as Doctor Lehman has had, in that we have had a higher mortality rate in hyperthyroidism in the Negro race. I have no exact figures, but I do know that in the white group we have not had a death during the past eight years, while in the colored group I believe we have had three deaths. Since there are fewer colored patients operated upon for hyperthyroidism, these figures seem significant.

My impression would be that this is not due to any condition inherent in the Negro race, but that it is connected with a state of malnutrition. With their general living conditions most of these people are in a state of chronic malnutrition; and while some are apparently well-nourished, it is probable that even these have a vitamin deficiency.

DR. JAMES D. RIVES (New Orleans, La.): The statistics quoted by Doctor Maes from New Orleans have been a constant reproach to us and to the Hospital for a period of years. They are the worst figures in the country, but we believe they serve a useful purpose in that we may be able to help teach surgeons what not to do in the handling of thyroid disease. There are several factors that have entered into that mortality; they are clear, and I think deserve to be emphasized at this time: First, a large number of these cases have been operated upon by surgeons who only occasionally perform a thyroidectomy, and not a few have been the first operated upon by that surgeon. We all know that the mortality of thyroid disease depends to a considerable extent on a high degree of technical skill. Second, in our part of the country there is a misunderstanding about the rationale of treatment; prolonged iodine administration has been the rule rather than the exception. I would like to practice where Doctor Lehman does, where the incidence of prolonged iodine therapy is only 30 per cent. Third, we have a very large Negro population; that is not a reason, naturally, except insofar as the hygiene and nutrition of the Negro is peculiar to the race. Fourth is the handling of the patient in the hospital after he is admitted.

In most hospitals, proper provision is made for segregating cases. It is true that we rarely put more than two hyperthyroid cases in one bed, but our wards are very crowded. I lost one in a nine-bed ward that was housing 21 patients. The danger of improper handling is one thing we all know, but it is a thing we may forget if we do not see the results before our very eyes. We do not expect this condition to continue indefinitely in New Orleans, but at present it is necessary that we operate our hospital for two years with half the requisite number of beds, and the results have been reflected in the thyroid mortality more than in any other disease. We have had, on our own service, five patients die of thyroid crises during a period of preoperative preparation; at no time in their progress did we feel there was any chance that operation would have been successful. We did not give up the idea of operative treatment early enough to try other methods.

One of the conditions that I think deserves emphasis is the fact that five of the seven cases who have died in a thyroid crisis without operation on our own service, have died during the months of July and August. Last August,

when we lost four, the average maximum daily temperature was over 90° F., and the humidity about 85 per cent. The loss of body heat is interfered with much more in our community than in most others.

Dr. T. C. Davison (Atlanta, Ga.): I would like to say just a word with reference to the intravenous treatment of hyperthyroidism. I wonder if Doctor Lehman has used iodine in that way. This method of treatment is especially applicable when "time is the essence of the contract" and one wishes to shorten the preoperative stage.

A patient comes into the hospital, and on the first morning a basal metabolic determination is made, then 1 Gm. of sodium iodide is given intravenously that day, 2 Gm. the next day and the basal metabolic reading is repeated on the third morning. It is usually found to be reduced approximately one-half in toxic cases; then two more grams of sodium iodide are given that day, and on the morning of the fourth day the patient is operated upon. This is not an iron-clad rule, but one can materially shorten the hospital stay in toxic cases by employing this treatment.

In recent years I have been removing more gland in toxic cases and I have better results and fewer recurrences. I formerly removed 75 per cent of the thyroid gland and now in severely toxic cases I remove 90 to 98 per cent of the gland.

Dr. Robert S. Dinsmore (Cleveland, Ohio): Doctor Lehman's observations about exophthalmic goiter in Negroes have been interesting to note, as we have seen it only infrequently in colored people. At one time we had a series of cases in which it had occurred in colored ministers. Doctor Crile, at the time, had an interesting unpublished theory as to the etiology of the disease in this group.

Doctor Lehman mentioned the preoperative use of iodine as given by the family physician. Without entering into the time-worn discussion about this problem, I wish to make one point, which is, that in instances where these patients have had iodine over a long period of time it is far better to maintain this dosage rather than to attempt any changes. Apparently they have established a maintenance dose, and almost invariably where this has been changed we have prolonged the preoperative period of these patients.

RESULTS OF THYROIDECTOMY IN HYPERTHYROIDISM ASSOCIATED WITH NEUROCIRCULATORY ASTHENIA

WILLIAM H. PRIOLEAU, M.D.
CHARLESTON, S. C.

EMPHASIS is so generally placed upon the differentiation of hyperthyroidism from neurocirculatory asthenia that, not uncommonly, it is lost sight of that these conditions may occur together. Only a few cursory allusions to this fact are found in the literature. Crile[1] makes mention of it and further states that the presence of neurocirculatory asthenia may not be recognized until after thyroidectomy; also that thyroidectomy has no effect upon the neurocirculatory asthenia. Craig and White,[2] in an analysis of a series of cases of neurocirculatory asthenia, report one case associated with "thyrotoxicosis" and one with an "adenoma of the thyroid"—in both of which, after the eradication of the thyroid disease, the neurocirculatory asthenia remained. Rienhoff[3] states that hyperthyroidism may be, and often is, engrafted upon a condition of autonomic imbalance, psychoneuroses, neurocirculatory asthenia, or some other allied functional disorder. The authors mentioned give no case reports or detailed consideration of the subject.

In both hyperthyroidism and neurocirculatory asthenia,[4] the sympathetic nervous system is fundamentally affected and accordingly the symptomatology of the two diseases is similar in some respects. Hyperthyroidism is characterized by nervousness of an active type[5]—a drive—the result of excessive stimulation by the overactive thyroid gland. In addition there is generally an increased basal metabolism. The resultant clinical picture is that of tachycardia, palpitation, excessive nervousness, ease of fatigue and loss of weight. Neurocirculatory asthenia is a variable clinical syndrome of undetermined etiology. There is considerable evidence that it is of developmental or constitutional origin, while some cases apparently have followed influenza, severe strain, *etc.* In it there is an instability of the sympathetic nervous system. Common symptoms are tachycardia and palpitation of varying degree in contradistinction to their persistency in hyperthyroidism, nervousness of a listless type, emotional instability, ease of fatigue, sighing, cold moist hands and feet, and neurotic symptoms with reference particularly to the gastro-intestinal tract. The nutrition is not affected; however, the basal metabolic rate not infrequently is elevated on occasions—explained by McCullagh[6] as being due to the very unstable state of the sympathetic nervous system. Further differentiation will not be considered here, but it should be emphasized that not infrequently a diagnosis is not to be reached except after a period of observation, and in an occasional case not even then.

In the cases in which the two diseases coexist, there are characteristic features of each, and accordingly a diagnosis of only one is insufficient to explain the clinical picture. The symptoms vary as in each of the two diseases and ac-

cording to the predominance of one or the other. In a series of 225 consecutive thyroidectomies, the author has encountered three cases which definitely belong in this category. There were other cases less pronounced and accordingly of a more uncertain nature.

CASE REPORTS

Case 1.—The patient, white, female, age 20, unmarried, was first seen July 1, 1936. Of a nervous family, she had never enjoyed good health and had been a semi-invalid ever since she could remember. Four or five years previously, she noticed an enlargement of her neck for which she had taken iodine at times. During the past year, her nervousness and tenseness have been greatly increased. She has had frequent attacks of palpitation and dyspnea upon climbing one flight of stairs. She has choking sensations and some difficulty in swallowing. During the past two years her menstrual periods have been coming at longer intervals. She has always had trouble with her eyes.

Physical Examination revealed a nervous, restless, normally developed, moderately well nourished young woman. Weight 116 pounds; pulse varying from 120 to 90; blood pressure 140/90; temperature 99.2° F. There was a pronounced congenital muscle imbalance of the eyes, the left eye being depended upon for vision. The thyroid gland was moderately enlarged, nodular and firm. The heart was not enlarged, there were no murmurs; the precordial beat was forceful. There were internal hemorrhoids of moderate size. B.M.R. plus 19 per cent. Blood cholesterol 162 mg. per 100 cc. *Clinical Diagnosis:* Nodular goiter with mild pressure symptoms and hyperthyroidism, superimposed upon a condition of neurocirculatory asthenia.

October 27, 1937: A conservative, subtotal thyroidectomy was performed.

Pathologic Examination.—Gross: Two pieces of thyroid gland, each about 5x3x2 cm., and a smaller piece, aggregating 33 Gm. in weight. They are well lobulated and the acinar markings are prominent on cut surface. *Microscopic:* Acini generally enlarged and distended with smooth colloid; epithelium usually flat, low cuboidal in smaller acini; no definite framework or lymphoid increase; vessels generally full of blood. *Pathologic Diagnosis:* Nodular goiter of the colloid type.

Subsequent Course.—During the few months following the operation, the condition of the patient was variable—periods of improvement alternating with severe nervousness and palpitation.

March 22, 1938, five months after operation: Weight 114 pounds, pulse 130 to 90. Clinically a normal thyroid balance. General improvement pronounced, though still has periods of nervousness. Now seldom conscious of heart. Able to climb stairs and do housework for first time in years.

July 13, 1938, nine months after operation: Pulse 120 to 80. Occasional ectopic beat. B.M.R. plus 13 per cent. Blood cholesterol 132 mg. per 100 cc. Leading very active life, such as was not possible before operation. Not tense, but irritable at times.

September 22, 1938, 11 months after operation: Pulse 112 to 88. Sinus arhythmia 2 plus. Condition as on previous examination.

Case 2.—The patient, white, female, age 19, college student, was first seen, June 22, 1937. Mother nervous. Father thinks that his pulse has always been rapid. Two years previously, a school physician told the patient that her pulse was too fast. However, she had not become aware of it until six months ago, at which time her B.M.R. was plus 21 per cent. During the past six months, she has been nervous and irritable, conscious of palpitation, has had attacks of indigestion and vertigo after lying down.

Physical Examination.—The patient was a well developed, moderately well nourished, and moderately nervous young woman, with a staring expression. The thyroid gland was diffusely enlarged and moderately firm and bosselated. The heart was not enlarged; the precordial impulse was forceful. There was a questionable systolic murmur

at the apex. The B.M.R. was plus 45 per cent. *Clinical Impression:* Hyperthyroidism superimposed upon a condition of neurocirculatory asthenia.

July 18, 1937: A conservative subtotal thyroidectomy was performed.

Pathologic Examination.—Gross: Three pieces of thyroid tissue, two 4x2x1 cm., one 3x1x1 cm., aggregating 18 Gm. Finely lobulated and of coarse, translucent granularity on section. *Microscopic:* Acini on average larger than normal, varying considerably in size, the larger having flattened epithelium, the smaller low cuboidal, all filled with smooth colloid. No definite increase of framework or lymphoid tissue. Vessels generally filled with blood. *Pathologic Diagnosis:* Diffuse enlargement, colloid type.

Subsequent Course.—December 12, 1937, five months after operation: Weight 149 pounds (gain of 36 pounds since operation). Pulse 120 to 90. Much less nervous; heart much quieter. Pulse rate less rapid at end of visit; much greater endurance. Attacks of indigestion less severe and less frequent. Impression: Normal thyroid balance; remaining symptoms due to neurocirculatory asthenia.

July 2, 1938, one year after operation: Weight 133 pounds; pulse 130 to 100; blood pressure 128/80. No finger tremor. Eye movements normal. Still nervous, but much less than before operation. Now has palpitation only on excitement or exertion. Able to do a full day's work, though tired at the end, whereas formerly able to work only half a day. Complains of empty feeling a few hours after eating. Blood cholesterol 132. B.M.R. plus 13 per cent.

Case 3.—The patient, white, female, age 16, was first seen in September, 1930. Eight months previously, she noticed an enlargement of her neck. At that time, she became very nervous, cried easily, and had attacks of palpitation.

Physical Examination.—The patient was small, normally developed, moderately well nourished and nervous. The thyroid gland was diffusely enlarged and of firm consistency. The heart was not enlarged, the precordial impulse was forceful, there were no murmurs. Pulse 110; blood pressure 124/68. B.M.R. plus 23 per cent (Benedict's for girl). *Clinical Diagnosis:* Hyperthyroidism; atypical type with heart symptoms predominating.

October 3, 1930: A subtotal thyroidectomy was performed.

Pathologic Examination.—Gross: Two masses of thyroid tissue, moderately tough, slightly lobulated, light beefy color, weighing 14 Gm. *Microscopic:* Acini generally enlarged; colloid content increased, smooth; epithelium low cuboidal to flat; no definite framework or lymphoid increase; vessels not conspicuous. *Pathologic Diagnosis:* Diffuse enlargement, colloid type.

Subsequent Course.—Three months after operation, she was less nervous, had greater endurance; the tachycardia and palpitation persisted but to a less degree. June, 1931, nine months after operation: Heart action rapid and forceful. Roentgenologic examination reveals no enlargement of heart. Patient appeared to have a normal thyroid balance. February, 1933, two and one-half years after operation: Feeling well; no heart attacks. Pulse 120, but slow at times. August, 1938, eight years after operation: Returned for examination by request. Working hard. Pulse 102, apical impulse forceful. Cardiologist unable to detect organic heart disease. Only seldom conscious of heart. Appears to have normal thyroid balance. B.M.R. minus 7 per cent. Blood cholesterol 230 mg. per 100 cc.—possibly accounted for by removal of most of her ovarian tissue in a recent operation, though still menstruating.

COMMENT.—The three cases reported are all in young women. In all there was a definite period of onset of the present illness—apparently the time at which the hyperthyroidism developed. Indicative of the presence of neurocirculatory asthenia, these patients had cardiac overactivity and ease of fatigue out of proportion to the degree of hyperthyroidism as determined by the other clinical and laboratory data. All three patients were subjected to a conserva-

tive subtotal thyroidectomy. In one, the gland was nodular, in two, it was diffusely enlarged, in none was there microscopic evidence of active hyperplasia, a condition probably accounted for by the use of iodine. All three patients developed, after a few months and still had at the time of last examination, a state of normal thyroid balance as judged by general appearance, skin, hair, mental alertness, absence of abdominal bloating and muscle and joint stiffness, and the basal metabolic rate. Notwithstanding this the symptoms of tachycardia, palpitation, ease of fatigue and nervousness persisted, though to a much less degree. The persistency of these symptoms cannot be accounted for by permanent damage resulting from a very severe or prolonged hyperthyroidism,[7] as in these cases the hyperthyroidism was of a mild degree and not of long duration. Nor can the improvement be attributed to an induced hypothyroidism, in view of the evidence of a normal thyroid balance, and also the well recognized fact that hypothyroidism is not well tolerated by a patient with neurocirculatory asthenia. In none of the three patients could the presence of organic heart disease be determined.

The improvement has been mostly from a subjective standpoint. Previously, these patients were practically always conscious of their heart and were able to do only a half day's work. Now they are conscious of their heart only on occasions and are able to do a full day's work. In Case 3, in which the heart symptoms were most pronounced, there has been continued improvement, especially during the past two years—which is most likely accounted for by an abatement of the neurocirculatory asthenia.

In these cases, the treatment has been directed only toward relief of the hyperthyroidism. Accordingly, all improvement can reasonably be attributed to it, except in Case 3, in which there has been further subsidence of the cardiac symptoms taking place several years after the establishment of a normal thyroid balance. While these patients are not well, due to the continued presence of the neurocirculatory asthenia, their improvement has been such as to fully justify their having been subjected to an operation for relief of the coexisting hyperthyroidism. In such cases, as in others, the diagnosis of hyperthyroidism is, in itself, sufficient indication for thyroidectomy[5]—barring definite contraindications. In making the diagnosis of hyperthyroidism associated with neurocirculatory asthenia, the final decision must rest upon a careful history and physical examination, often followed by a period of observation. Clinical experience is to be relied upon more than laboratory tests. Unless great care is exercised, patients with neurocirculatory asthenia alone will be subjected to thyroidectomy, often with resultant harm.

CONCLUSIONS

Hyperthyroidism and neurocirculatory asthenia may coexist, in which case there are characteristic features of each disease. In these cases, thyroidectomy relieves the hyperthyroidism; the symptoms of neurocirculatory asthenia persist—the patient, however, is greatly improved.

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DISCUSSION.—DR. ROBERT S. DINSMORE (Cleveland, Ohio): Doctor Prioleau has presented a group of patients that are always confusing to the surgeon. The difficulty arises as to whether they have neurocirculatory asthenia or hyperthyroidism, or a combination of the two; and, if the latter, it may be difficult to determine whether the asthenia or the hyperthyroidism is the predominating syndrome. On the whole, operations upon this group of patients have been disappointing. The second case which he reported was the most clear-cut of the group. Frequently, these cases will be markedly improved for four to six months and then the asthenic symptoms are apt to recur. One of the striking characteristics about these patients is that they do get rid of the heart consciousness which is a prominent symptom before operation. The same patients may have a persistent tachycardia but the subjective symptoms have been relieved. The differentiation of the two diseases primarily may be difficult. One clinical observation has been of help to me, which is that the patient in the hospital should have two-hour pulse readings. If the pulse comes down quickly and strikes a base line, one may be reasonably sure one is not dealing with hyperthyroidism.

DR. FRANK H. LAHEY (Boston, Mass.): As regards the idea of operating upon patients with neurocirculatory asthenia, I am always at a loss to know how to disagree with a man who is younger than I am, and I wish particularly to disagree kindly. When such a term as neurasthenia is applied to conditions, it means, I believe, that they do not know what it is or what to do about it. In spite of this, I think the term is as good as any that can be applied to the condition.

It is extremely important, I think, to be sure that these borderline patients, in whom the distinction has not been made as to whether they are of thyroid or neurocirculatory origin, are not operated upon until it is established that they are definitely cases of hyperthyroidism. I am sure that if they are not, surgery is of little or no value and even harmful. Time and repeated observation will establish the fact that a patient has hyperthyroidism or does not have it. If frank hyperthyroidism does not appear, so that it can be clinically diagnosed within a few months, after repeated observation, it is usually not present.

DR. WILLIAM H. PRIOLEAU (Charleston, S. C., in closing): As regards Doctor Lahey's discussion, I appreciate the spirit in which it was given. I expected it, I won't say from him, but from somebody. I fully realize that at best the treatment of this group of cases is unsatisfactory. To be more explicit, we

use the term "neurocirculatory asthenia" loosely, inasmuch as it does not represent a clinical entity. However, there is no reason why a condition of hyperthyroidism should not be superimposed upon it. Due to only a moderate elevation of the basal metabolic rate, two of the cases reported may be open to question, but not so the third. As regards the basal metabolic rate in the diagnosis of hyperthyroidism, it is my experience that hyperthyroidism may exist with the basal metabolic rate not elevated above the upper limits of normal. On the other hand, operation solely upon the basis of an increased basal metabolic rate would result in a number of unnecessary thyroidectomies. Similar experiences are reported by other observers. The absence of microscopic evidence of hyperplasia does not disprove that hyperthyroidism did not exist. In the cases reported, the disease was of a mild type, and the use of iodine generally results in a remission.

TRAUMATIC ARTERIOVENOUS ANEURYSMS OF THE GREAT VESSELS OF THE NECK

OBSERVATIONS UPON SEVEN CASES

JAMES M. MASON, M.D.

BIRMINGHAM, ALA.

INJURIES to the great vessels of the neck always present problems of interest and gravity. The anatomic relations of these vessels are important. Pressure from hematomata or from expanding aneurysmal tumors may interfere with physiologic function, and accidents during operative attack may be followed by disastrous consequences.

Since January, 1932, seven traumatic arteriovenous aneurysms involving the great vessels of the neck have come under my observation. All resulted from gunshot or stab wounds. The following vessels were involved: Right subclavian vessels, third portion, two cases; left subclavian vessels, third portion, three cases; left subclavian artery and left innominate vein, one case; left external carotid artery and internal jugular vein, one case.

Two patients refused operation and left the hospital shortly after examination. One patient died on the fourth day after injury, before any surgical measures were undertaken for his relief. Four patients were operated upon —one by multiple ligation and excision of the fistula, and three by transvenous arteriorrhaphy. All the operative cases recovered and three were cured. One patient improved following excision of a fistula between the left subclavian in its first portion and the left innominate vein, but died six years later from the effects of an additional fistula between the common carotid and left innominate vein, which had not been discovered at the time of operation but which was revealed at autopsy.

The two cases which refused operation were respectively of 3 and 14 years' standing. In the former case, marked cardiac degeneration was present; in the latter, the terminal stage of decompensation was approaching and the patient died within a few weeks after leaving the hospital. The details of the case that died without operation four days after injury have been reported,[7] together with autopsy findings.

Papers describing the cardiovascular or systemic effects of arteriovenous communications were presented by Matas at the meeting of the Southern Surgical Association in 1923 and by Reid at the meeting in 1931. In addition, many communications have appeared in the literature confirming the fact that, in arteriovenous communications between vessels of larger caliber, serious cardiac degeneration will follow if the condition remains long uncorrected. The main damage to the heart results from the quick shunting of large quantities of arterial blood directly into the vein and thence to the heart. It will not be necessary to dwell on this, further than to briefly discuss certain phases.

It has been very well established that the larger the vessels and the closer

the fistula to the heart, the more rapid and severe will be the degenerative effect upon the heart muscle. Nevertheless, in reviewing a large number of reports, and in my own small series, there has been great variation in the time of appearance as well as in the rapidity of progress of degenerative heart changes.

I have devoted some time to speculation concerning possible causes for these variations. If we except the case in which the communication was in the first portion of the left subclavian artery, the fistulae in my series did not differ materially in size or in proximity to the heart. The two cases of long standing showed marked cardiac changes, though we do not know, definitely, how early they appeared.

In the remaining five cases, we had the opportunity, at operation or autopsy, of examining the condition of the vessels at the site of the fistula and adjacent thereto. In Cases 3 and 4, cardiac changes were noted early. One patient, Case 3, died on the fourth day from decompensation; another, Case 4, became bedridden from the same cause within a month. The third patient, Case 5, gave a positive Branham's sign and exhibited characteristic blood pressure phenomena on closing the fistula by compression, within a week after his injury. He was operated upon 27 days after his injury, and had shown no clinical signs of heart involvement. However, during this time he was confined to bed by other complicating injuries and the minimum tax was placed on his circulatory system. Branham's sign and the blood pressure phenomena are considered indicative of cardiovascular response to the presence of an arteriovenous communication.

In these three cases, the fistulae were direct communications between the artery and vein, without varicosities, thrombosis, or other evidences of obstruction to the direct return of arterial blood through the fistula to the vena cava.

Cases 6 and 7 were under continuous observation from the time of injury to that of operation: four months later, in one case, and six months later, in the other. Cardiac symptoms were anticipated and repeatedly searched for, but none appeared. The fistulae were inaccessible to closure by compression, hence Branham's sign and blood pressure phenomena could not be elicited. One patient had a pulse rate which was persistently above normal during the first month, and this may have been significant, as it remained normal thereafter. Soon after his injury, he developed phlebitis in the veins about the fistula, which extended down the arm, but this had undergone resolution at the time of operation.

In both of these cases, we found at operation that large venous sacculations and varicosities began at the immediate location of the fistula and extended well down to the anterior axillary border.

The location of these varices, beginning at the very point of the orifice of the communication, would seem to indicate that, at the site of the fistula, there was obstruction to the return flow of venous blood. The obstruction

may have been sufficient, also, to interfere with free entry of arterial blood into the vein through the fistula. If these inferences are correct, they give a possible explanation of the lack of development of cardiac symptoms during the months which preceded operation.

They are interesting, too, in that they tend to confirm the clinical and experimental observations of other workers in this field. Holman and Stultz found in arteriovenous aneurysms experimentally produced that, in some instances, cardiac lesions failed to develop either early or late. This failure was attributed to the formation of a thrombus in the vein proximal to the fistula. Organization of the clot prevented reflux of blood toward the vena cava, and dilatation of the right heart did not occur. Tixier and Arnulf,[1] in reporting a case of very early cardiac decompensation, occurring on the fifth day after an injury, which resulted in an arteriovenous fistula of the femoral vessels, say: "Theoretically, if we accept the experimental work of Holman and Stultz, it might be possible to begin treatment by the application of a ligature on the vein some distance proximal to the fistula. The experiments of these authors show that by this simple procedure one may stop cardiac disorders." Holman[2] reports one clinical case where "simple closure of the vein proximal to the fistula obliterated the thrill of an arteriovenous aneurysm and caused the pulse to drop from 112 to 84." Harvey Stone[3] had a similar experience, where immediate improvement followed ligation of the vein. Matas[4] comments favorably upon the "possibilities of proximal ligation of the vein alone in severe decompensation, to stop the strain on the heart and permit a more complete operation at a later date, when the collateral circulation is fully established."

While these writers discuss only ligation and thrombosis of the vein proximal to the fistula, their observations tend to establish the principle that cardiac dilatation will be prevented, delayed, or arrested, for a time, if the free flow of arterial blood into the vena cava is interfered with, either by accidental thrombosis or intentional ligation of the vein proximal to the fistula.

In my cases, while the varicosities were on the distal side of the fistula, the obstruction to the venous flow was at the site of the fistula, and it is reasonable to suppose that it resulted in partial occlusion of the fistula itself, and thereby materially interfered with the return flow of arterial blood to the vena cava in a manner similar to the proximal ligations above referred to.

Operative Technic.—Special attention should be given to certain important details in operations upon arteriovenous fistulae in the vessels of the neck. I have employed both local and general anesthesia with equal satisfaction. If the heart is seriously affected, local anesthesia is safer, as the operations are essentially tedious and prolonged.

The surgeon has the choice of absorbable or nonabsorbable suture material in the general conduct of his operation, but silk must be insisted on for arterial ligations and sutures.

In operations on the subclavian vessels, it will often be necessary to

divide and retract the clavicle. This may afterwards be excised or replaced and wired. I have replaced and wired it in two instances, and have removed the inner half in one instance without apparent loss of strength or impairment of function of the arm. In reviewing a collection of 19 operations, we find it stated that the clavicle was excised, wholly or in part, in eight instances, and was replaced and wired in three instances. In a case reported by Gilcreest, in which a two-stage operation was performed, the clavicle was excised subperiosteally at the first operation and had reformed at the second operation, necessitating another excision.

Hemostasis is an important problem, since, in the region under discussion, a tourniquet cannot be employed. Before attacking an arteriovenous fistula directly, provisional ligatures or clamps should be placed on the artery and vein above and below the fistulous communication. In some instances quadruple ligation cannot be accomplished. However, it is generally possible to expose the artery on the proximal side of the fistula and obtain control of the source of the most active circulation. It may then be possible to limit venous hemorrhage by pressure and other methods. The most ingenious method of occluding the opening of inaccessible fistulae is that devised by Reid,[8] in which he ligates and frees the vein some distance below the fistula and twists it until the opening is occluded. The vein is then sutured to adjacent soft tissues to prevent it from becoming untwisted.

In recent injuries, except under certain conditions where the urgency of the situation demands immediate operation, regardless of potential secondary peripheral circulatory incompetency, it is advisable, as has been pointed out by Matas, Reid and others, to wait from three to six months before undertaking curative operations. The advantages of delay have been set forth in other communications and will not be repeated.

Numerous observations have shown that an adequate collateral circulation is rapidly established in most instances, making quadruple ligation and excision safe in delayed cases. In cases of long standing this is the method of choice, since the degenerative changes which are likely to have taken place in the arterial wall make it probable that aneurysmal dilatation will follow attempts at arteriorrhaphy. In the neck, and in other regions, I have practiced quadruple ligation and excision without interference with peripheral circulation.

In healthy young patients, whose only vascular pathology is a recent wound, it has seemed well to preserve the main arterial supply to distal parts by closing the arterial wound by transvenous arteriorrhaphy. Arteriorraphy has sometimes been followed by thrombosis with occlusion of the lumen of the vessel, but in the three instances in which I have closed the fistula in this manner, the peripheral circulation has been preserved. It is a less formidable operation than excision, and may be safely employed in selected cases. This operation, as applied to the subclavian vessels, was first performed by Matas,[5] in 1921.

Ligation of the left subclavian artery in the first portion is extremely difficult. My experiences in accomplishing it in Case 4 were reported in detail in a paper presented at the meeting of the Southern Surgical Association in 1932. It was necessary to resect the clavicle, the anterior half of the first rib, and the outer half of the manubrium sterni, in order to reach the artery near its origin from the aorta. The operation of multiple ligation and excision of fistula was performed in two stages, under local anesthesia, without mishap.

In the external carotid-internal jugular fistula (Case 5), dissection of the neck and repair of the fistula by lateral arteriorrhaphy, under local anesthesia, was attended with no difficulty other than that due to the high location of the fistula which made hemostasis rather hazardous. A provisional ligature was placed around the artery proximal to the fistula. The vein was tied below the fistula and clamped above it. The fistula was exposed and hemorrhage was controlled by digital compression until sutures could be passed and the vein above the communication closed by deep sutures.

Two cases involving the third portion of the subclavian artery were operated upon, under general anesthesia, through an incision which gave adequate exposure of the fistulous communication and permitted arteriorrhaphy to be carried out without difficulty. The steps are as follows:

OPERATIVE STEPS IN TRANSVENOUS SUTURE OF AN ARTERIOVENOUS FISTULA IN THE THIRD PORTION OF THE SUBCLAVIAN ARTERY AND VEIN

(1) Skin incision: Beginning at upper border of the thyroid cartilage over outer border of sternomastoid muscle, down to its clavicular attachment, across the upper surface of the clavicle to its outer third, thence downward and outward to the anterior axillary border.

(2) Dissection and retraction of skin flaps; retraction inward of sternomastoid muscle to expose scalenus anticus; retraction of this muscle inward to permit provisional ligation of subclavian artery.

(3) Division of the clavicle; division of the clavicular attachment of the pectoralis major; division of subclavius and of tendon of pectoralis minor.

(4) Retraction of the clavicle to permit proximal ligation of the subclavian vein. It is important to make this proximal ligation before opening into the aneurysm, as fatal air embolism has followed incision into the arteriovenous aneurysm before the vein was closed proximal to the fistula.[6]

(5) Freeing the subclavian artery and vein well beyond the wounds in the vessels with provisional ligation of the artery. Freeing any large mass of varicosities and ligating the vein well below them.

(6) Incision of vein to expose the site of the fistula; transvenous suture of wound in artery; removal of provisional ligatures to test the line of suture for leakage; excision of redundant sac and suturing the remaining portion over the site of the arteriorrhaphy.

(7) Reconstruction of the wound in the proper anatomic planes; wire or remove the clavicle as may be determined upon; skin closure without drainage.

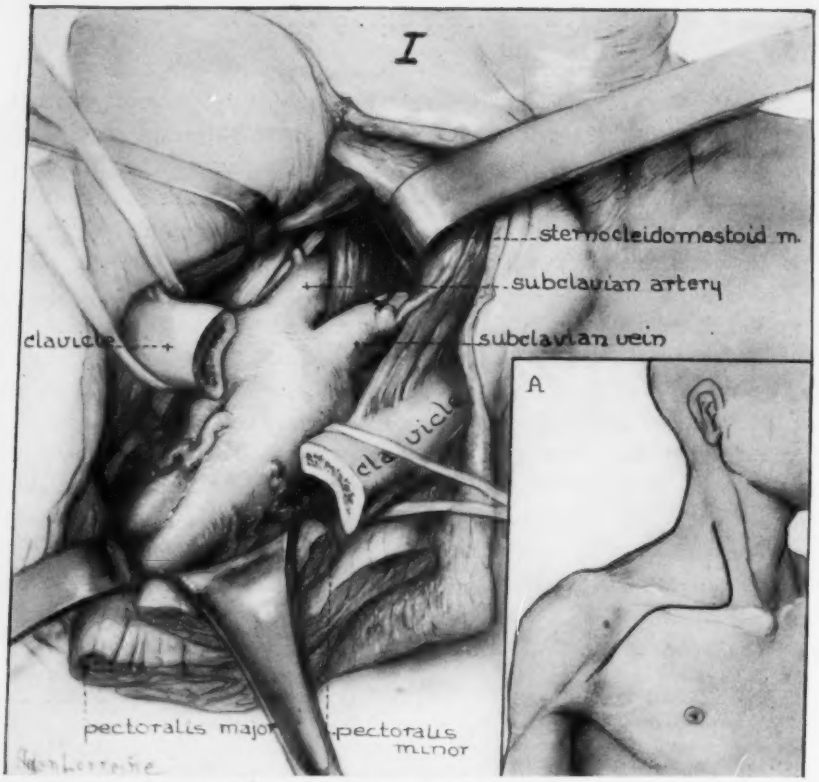

FIG. 1.—(A) Skin incision. (I) Retraction of flaps; retraction of sternomastoid muscle to expose scalenus anticus muscle, and artery and vein in front and behind this muscle; division of muscles and of clavicle to expose aneurysm and vessels above and below fistula.

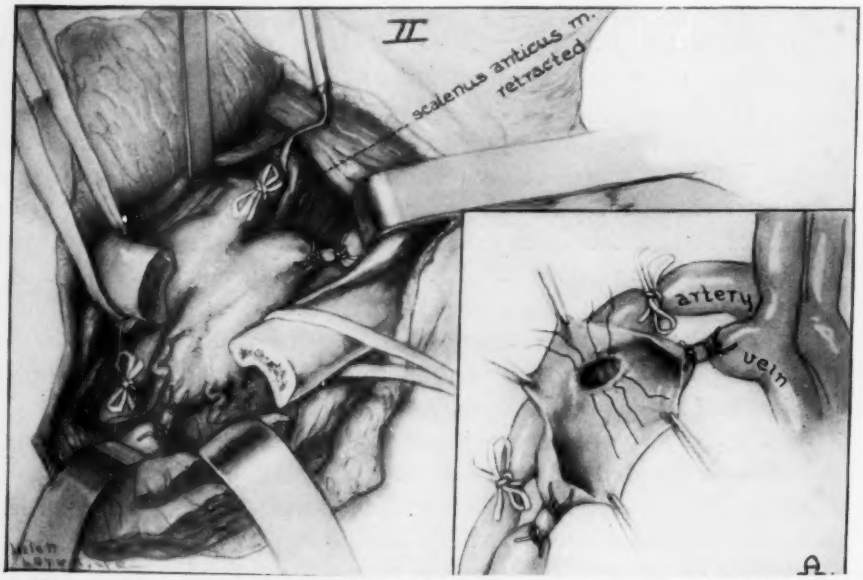

FIG. 2.—(II) Provisional ligatures have been placed on artery above and below fistula. The vein has been doubly ligated above and below the fistula. (A) The fistulous orifice has been exposed by incision of the vein, and sutures have been placed in arterial wall.

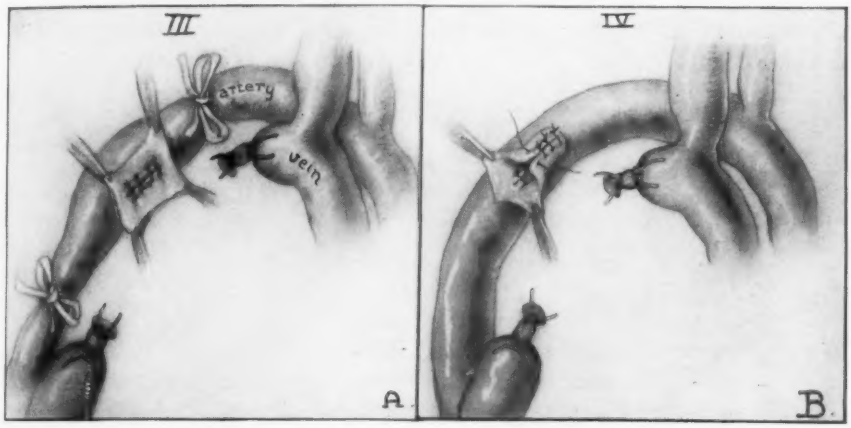

FIG. 3. FIG. 4.

FIG. 3.—(A) The redundant venous sac has been trimmed away and the arterial wound has been closed by interrupted sutures of black silk.

FIG. 4.—(B) The provisional ligatures have been removed from the artery, and, after being assured that there is no leakage, the arterial suture line is reinforced by suturing over it the remains of the venous sac.

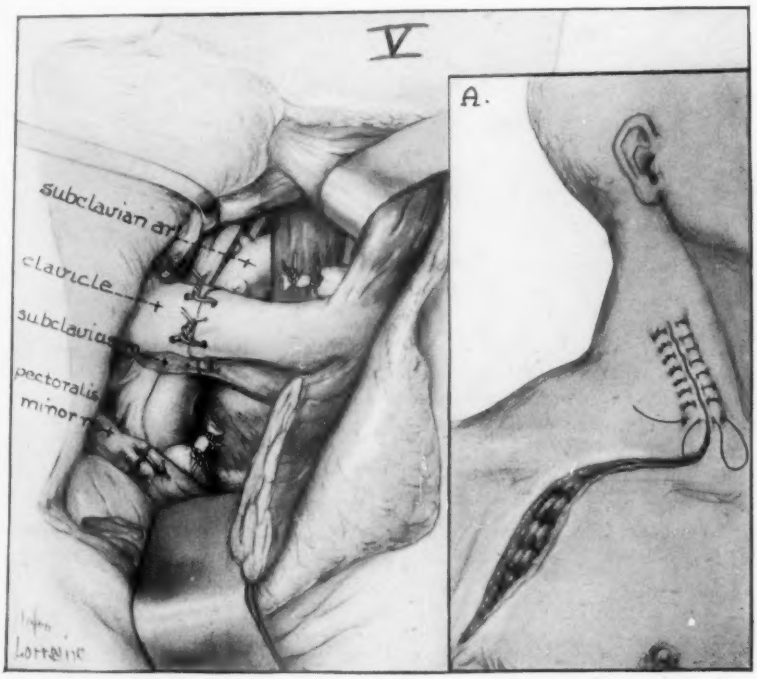

FIG. 5.—(V) The provisional ligatures have been removed from the subclavian artery; each muscular and fascial plane has been reconstructed; the clavicle has been wired. (A) The skin closure is shown.

CASE REPORTS

Case 1.—Hillman Hosp. No. 64706: *Arteriovenous aneurysm left subclavian vessels, following stab wound left supraclavicular fossa three years previously. Refused operation.*

T. E., colored, male, age 29, examined December 26, 1933, complaining of dizziness

and dyspnea on exertion. On October 21, 1930, he was stabbed in the left supraclavicular fossa and was treated in Hillman Hospital for a period of ten days. When discharged, his wound had healed, and the record does not show any mention of an arteriovenous fistula. He continued his work as a chauffeur and some months later began to suffer from dizziness and dyspnea. About two years after the injury, he was examined in another clinic and the presence of the arteriovenous aneurysm was discovered.

Physical Examination showed the typical signs of arteriovenous aneurysm, together with marked enlargement of the heart and mitral and aortic murmurs. The location of the wound makes it possible that the first portion of the artery may be the site of the fistula, but this cannot be determined without operation. He refused operation and has been lost sight of.[9]

Case 2.—Hillman Hosp. No. 121614: *Arteriovenous aneurysm right subclavian vessels, following pistol wound right second interspace below middle of clavicle, 14 years previously. Refused operation.*

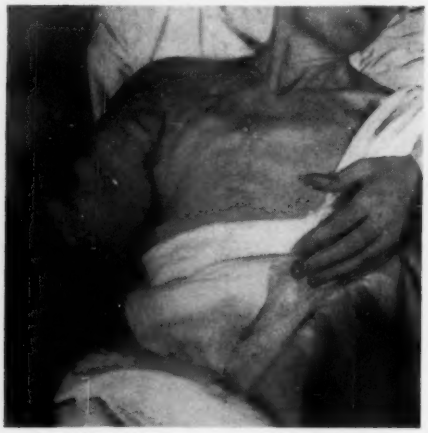

FIG. 6.—Case 2: Arteriovenous fistula, right subclavian vessels; 14 years' standing. Shows enormous size which the varicosities may attain in neglected cases.

W. E., white, female, single, age 42, was admitted to Hillman Hospital, April 28, 1938; refused treatment, and was discharged April 30, 1938. She died a short time later at her home. The patient received a bullet wound in the left second interspace below the middle of the clavicle, 14 years ago. A few weeks later there was noticed swelling in the region of the shoulder with progressive enlargement of the veins of the neck, right chest, right axilla and right arm. This was accompanied by dyspnea, cardiac irregularity, palpitation, precordial pain, and loss of weight. Three days before admission she developed acute redness and swelling about the elbow, and on admission thrombophlebitis was present.

Physical Examination revealed thrill, bruit, cardiac dilatation, murmurs at base and apex, and enormous dilatation of veins of right side of neck, right arm, right chest and axilla. The temperature was elevated; and the arm in the region was acutely inflamed. The cardiothoracic ratio was 59 per cent. The patient refused to consider any operative efforts for her relief and left the hospital April 30, 1938 (Fig. 6).

Case 3.—Hillman Hosp. No. 110217: *Traumatic arteriovenous aneurysm left subclavian vessels, third portion; following stab wound in left suprascapular region. Death on fourth day.*

A colored male, laborer, married, age 29, was admitted to the hospital, October 10, 1936. Died October 14, 1936. This case is unusual on account of the rapidity with which degenerative heart changes took place, resulting in death on the fourth day after injury.[7]

Case 4.—Hillman Hosp. Nos. 72748 and 82721: *Traumatic arteriovenous aneurysm left subclavian artery, first portion, and left innominate vein; following stab wound below clavicle near the border of the sternum. Two-stage operation: Ligation left subclavian artery in first and third portions; left internal jugular, subclavian and innominate veins and excision of vessels in the ligated area. Operative recovery, with improvement of patient.*

The patient died six years later from a recurrence of symptoms of cardiac decompensation, and autopsy revealed an arteriovenous aneurysm of the left common carotid and the left innominate vein which had not been discovered at operation.

As improvement but not cure followed excision of the fistula between the left subclavian artery and left innominate vein, and since the cause of the recurring decompensa-

tion was not clear, the patient was kept under observation until her death, January 23, 1938, six years after her injury, January 17, 1932.

The following statement occurs in the previous report of this case: "The heart, though well compensating, still shows an aortic thrill and bruit as well as a soft double murmur at the mitral valve. I attribute these findings to the extensive damage which the heart sustained before the fistula was closed."

The operation of excision of the fistula was performed, May 3, 1932. She was discharged much improved, September 15, 1932. Her first readmission on account of decompensation was on June 6, 1933. Since that date she has been in and out of the hospital on numerous occasions, suffering from decompensation; and death finally took place, January 23, 1938. At autopsy a fistulous communication was found between the left common carotid artery very close to its origin from the aorta, and the left innominate vein. This had not been discovered at the time of operation. I have thought that it might have been possible that this was the only lesion present at the time of the operation upon the subclavian, but this could hardly have been the case.

From our present knowledge of arteriovenous communications, nothing which was done at the operation could have stilled the thrill and eliminated the bruit unless there had been a fistula between the subclavian artery and an adjacent vein. When the artery was ligated the thrill immediately disappeared. Both thrill and bruit slowly reappeared and were months in reaching the intensity which they formerly manifested. During this time, also, the patient showed marked clinical improvement. Other operators have reported the presence of two fistulae as a result of one accident, but fortunately they were in regions more accessible than in the present case and were not overlooked.[10]

Case 5.—Hillman Hosp. No. 99626: *Arteriovenous aneurysm of left external carotid artery and left internal jugular vein; following bullet wound of left side of face, July 4, 1935. Ligation and excision of vein. Arteriorrhaphy. Cure.*

Operation on twenty-seventh day after injury. Uneventful recovery. Circulation in temporal artery unimpaired.[9]

Case 6.—Hillman Hosp. No. 116700: *Arteriovenous aneurysm right subclavian vessels, third portion; following bullet wound of right thorax. Transvenous arteriorrhaphy. Cure.*

H. N. H., colored, female, married, age 27, was admitted to Hillman Hospital, August 20, 1937, suffering from a pistol wound of the right chest. The wound of entrance was in second interspace in midclavicular line. Exit near midline of the back opposite the fourth dorsal vertebra. The signs of arteriovenous aneurysm were detected within a few hours. The bullet had perforated the pleural cavity and a hemothorax was present, but disappeared without aspiration. She remained in the hospital until December 10, 1937, when she was allowed to go home for a few weeks upon promise to return for operation. The symptoms during the time of observation remained local, as manifested by thrill and bruit, and peripheral phenomena, as manifested by a persistently lower blood pressure on the involved side.

Physical Examination did not reveal any evidence of heart involvement, and teleroentgenograms taken, October 4, 1937, showed a cardiothoracic ratio of 50 per cent. On December 28, 1937, when she returned for operation, the ratio was 59 per cent.

December 29, 1937, under ether anesthesia, the fistula was closed by transvenous arteriorrhaphy. This resulted in complete cure. Convalescence was delayed by drainage from the silver wire sutures at the site of the division of the clavicle and persisted until the wire was removed. The radial pulse was unimpaired (Fig. 7).

Case 7.—Hillman Hosp. No. 116158: *Arteriovenous aneurysm left subclavian vessels, third portion; following gunshot wound left infraclavicular region. Transvenous arteriorrhaphy. Cure.*

D. C., colored, male, married, age 38, was admitted to Hillman Hospital, July 24, 1937, suffering from a recent pistol wound of left infraclavicular region. The bullet could be felt under the skin of the posterior aspect of the shoulder. A hematoma was

present, and there was also an injury to the brachial plexus, manifested by paralysis of the musculospiral nerve. The patient was carefully observed from day to day, but signs of arteriovenous aneurysm were not detected until the fourth day. After that date, thrill and bruit were continually present. He remained in the hospital until October 12, 1937, when he was allowed to go home to return for operation at a later date.

During this time, he did not develop any symptoms of cardiac involvement. The blood pressure on the two sides remained practically the same. The heart did not show any enlargement. Teleroentgenogram, September 7, 1937, showed cardiothoracic ratio of 40 per cent; and on January 5, 1938, when he returned for operation, the ratio was 46 per cent. On August 22, 1938, thrombophlebitis was noted in the veins of the arm, and this persisted for some time. The musculospiral injury was treated by massage, and a cock-up splint for the wrist. There was no improvement. For the first month after injury the pulse rate was persistently high, ranging from 80 to 120 daily. After this time it resumed a normal rate.

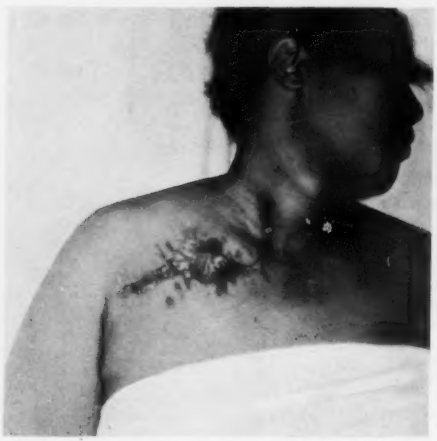

FIG. 7.—Case 6: Arteriovenous aneurysm of right subclavian vessels. Operative scar showing tendency to keloid formation.

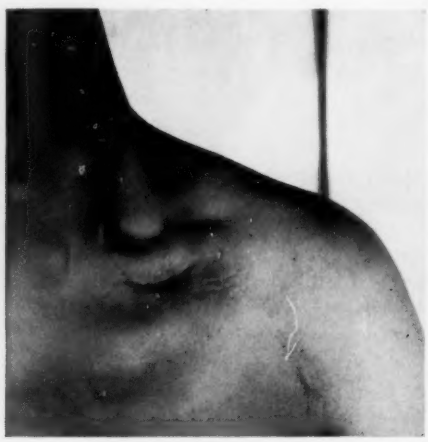

FIG. 8.—Case 7: Arteriovenous aneurysm of left subclavian vessels. Operative scar.

The patient was readmitted, January 4, 1938; and on January 6, 1938, under ether anesthesia, a transvenous arteriorrhaphy was performed. Cure. Radial pulse remained unimpaired. There was drainage from the site of the fractured clavicle, which continued until the wire sutures were removed. The musculospiral paralysis was unimproved. Such exploration of the brachial plexus as was possible at the time of operation failed to locate the nerve injury (Fig. 8).

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DISCUSSION.—DR. MONT R. REID (Cincinnati, Ohio): Doctor Mason has brought up so many important points that it is impossible to discuss them all. I am in entire accord with everything that he has said, but I do want to emphasize one or two points which he has made.

The first of these is the importance of bearing in mind that gunshot or stab wounds may produce more than one fistula. In a recent series we reported two such cases. The most common occurrence of more than one fistula is in situations where a large artery is accompanied by two venae comites; in such instances the injury may puncture both veins and artery and establish arteriovenous fistulae at the points of entrance and exit of the wound in the artery. In one such case recently, the surgeon operated and performed a transvenous closure of one fistula. Some four months later, it was necessary to perform a second operation and close the other fistula. It must be exceedingly rare to have two fistulae produced at separate levels involving different vessels, as Doctor Mason has reported in one of his cases.

The other point I wish to emphasize is that the mere size of the fistula cannot be regarded as a definite index of the amount of damage which will be done to the heart. The final test is how much actual increase in the cardiac output of blood is caused by the fistula. It would be advantageous if a satisfactory method for cardiac output determinations could be devised in humans with arteriovenous aneurysms. In this way we might be able to predict the probable rapidity of cardiac damage.

As has been indicated by Doctor Mason, there are many factors, such as occlusion of the proximal vein, thrombosis or the development of a large venous reservoir in the immediate vicinity of the fistula, which may actually reduce considerably the amount of blood actually returned to the heart, even though there is a large fistulous opening. In one case we found an extremely large communication directly between the subclavian artery and the large veins of the neck. This was a congenital lesion and, so far as I know, is about the only instance of a single, large congenital fistula excluding the ductus arteriosus. It had been present for 18 years, and yet there was no evidence of any cardiac enlargement or disability. There was not even a Branham's bradycardiac sign. The explanation in this case may be that, even though the fistula was large, the enormous dilatation of the involved veins led to some form of obstruction, which actually reduced the excess amount of blood which would normally have been returned to the heart if this large reservoir had not been present.

In another instance there was a fistula between the femoral vessels, no larger than the case just cited, but, after a period of eight years, there was extreme cardiac disability and decompensation. In this instance there was a relatively small reservoir of varicose veins in the region of the fistula. Finally, when acute cardiac disability does not result in a large fistula, such as is illustrated by some of the cases of Doctor Mason, and by the 19 cases reported by Pepper, in which aortic aneurysm ruptured into the vena cava with almost sudden death, it seems to me probable that factors come into play to produce eventual cardiac disability other than the actual amount of blood shunted back upon the heart. In long-standing fistulae it is unusual to see severe cardiac damage without a tremendous enlargement and dilatation of the proximal artery, which often extends all the way back to the heart.

In such cases, it has seemed to me reasonable to assume that alterations in the dynamics of the circulation on the arterial side may also be a definite factor in producing cardiac changes. As you know, Lewis has compared the effect of a large arteriovenous fistula with the effects of aortic insufficiency, believing that lowering of diastolic pressure produces decreased coronary blood flow and some myocardial ischemia.

DR. I. M. GAGE (New Orleans, La.): I wish to discuss only two phases of this subject: (1) The systemic effects of arteriovenous aneurysm, *i.e.*, cardiac dilatation in hypertrophy; and (2) the development of the collateral circulation distal to the arteriovenous fistula.

For years, Doctor Matas has repeatedly called our attention to the systemic effect of arteriovenous fistulae. He has demonstrated clinically that dilatation and hypertrophy of the heart follow the establishment of a traumatic arteriovenous aneurysm. Doctors Reid, Holman, Lewis, Herrmann and I have definitely proven that there is a cardiac dilatation following the experimental production of arteriovenous fistula in the dog. Herrmann and I, in 1927 to 1928, also demonstrated that not only is there a definite dilatation but a concomitant hypertrophy. We were fortunate in being able to show this, due to Herrmann's work on the heart weight, body weight ratio of the dog. Herrmann arrived at his averages of heart weight, body weight ratio from the study of 1,000 dogs. In all of our experimental arteriovenous aneurysms there was a definite increase in the heart weight, body weight ratio as well as dilatation. The experimental arteriovenous fistulae were made between the external jugular vein and the internal carotid artery, the femoral artery and vein, and the abdominal aorta and inferior vena cava.

The speaker then showed photographs of the comparison between a normal dog's heart and the dilated, hypertrophied heart of a dog that had an arteriovenous fistula between the internal carotid artery and the external jugular vein on the left side. The dog lived seven years following the establishment of the arteriovenous fistula. He was confined to a cage all of the time, and died suddenly of an acute dilatation of the heart. Herrmann believes that when the hypertrophy of the heart reaches a certain stage it becomes irreversible. However, this should be studied experimentally.

He performed complete pericardectomy upon ten dogs, in order to determine if the pericardium acted as an inhibiting membrane in dilatation of the heart following arteriovenous fistula. Following recovery from the pericardectomy, an arteriovenous aneurysm was made in each dog. We demonstrated that the pericardectomized heart dilated at the same rate as those with the pericardium intact. The pericardium has practically no inhibiting effect on the dilatation of the heart following experimental production of arteriovenous aneurysm.

As stated by Doctor Mason, the cardiac dilatation depends upon the size of the vessels involved, the size of the fistula, and its nearness to the heart. There is another factor that has not been mentioned and that is that the return of the excessive quantity of blood through the veins must not be impeded. If the return flow to the heart by way of the veins is impeded by narrowing of the veins proximal to the fistula, the systemic effects will not be marked. In fact, cardiac dilatation will not be present. I have had three cases recently that demonstrated this very conclusively. A male, age 67, had an arteriovenous aneurysm of the femoral vessel. The proximal vein was constricted by scar tissue which narrowed the vein to about two-thirds of its normal size. Although the aneurysm had been present for six years, roentgenographic and EKG. studies revealed a normal sized heart and no myocardial degeneration.

The second case, also, had no dilatation of the heart following a traumatic arteriovenous aneurysm of the femoral vessels of the right thigh. The patient, a male, age 32, had a traumatic arteriovenous aneurysm plus a large pseudo-arterial aneurysm, following a stab wound of the femoral artery and vein. Roentgenologic studies of the heart did not reveal any dilatation. The large arterial aneurysm acted as a safety valve in this case, and, therefore, the shunting of the arterial blood directly back through the veins to the heart was practically eliminated. The third case, demonstrating no dilatation of the heart, occurred in a young student, who had a stab wound of the anterior tibial artery and vein of the left leg. The duration of the aneurysm was eight years; the aneurysm was between the small anterior tibial artery and vein. The opening was very small and quite a distance from the heart. This case confirms the statement of Doctor Mason, *i.e.*, that the size of the vessel, the size of the opening, and the distance from the heart have a definite bearing on cardiac dilatation. Therefore, cardiac dilatation and hypertrophy of the heart do not occur in all arteriovenous aneurysms.

That a marked collateral circulation develops in the presence of an arteriovenous aneurysm has been proven both experimentally and clinically by many investigators. I believe the development of the collateral circulation depends to a great extent upon the size of the vessels involved as well as the size of the fistulous opening. Another factor which plays a very definite part in the development of the collateral circulation is constriction of the artery by scar tissue just distal to the arteriovenous fistula. Therefore, the collateral circulation depends upon interference of the circulation through the main arteries beyond the fistulous opening. The arteriovenous fistulous opening itself corresponds to a dam at the site of the fistula. This is proven by a diminution in the size of the main artery distal to the fistula and a dilatation of the artery on the proximal side to the aneurysm. Therefore, if there is a small opening between artery and vein, a minimum amount of arterial blood is shunted into the vein, with the result that the blood supply distal to the arteriovenous fistula is maintained almost entirely by the main arterial stem. If the fistula between artery and vein is large, a maximum amount of arterial blood is shunted from the artery into the vein; very little arterial blood reaches the distal part through the main arterial channels; therefore, a collateral circulation must develop to maintain adequate circulation of the part distal to the aneurysm. The development of the collateral circulation is progressive in character. The following case is reported to demonstrate the extensive collateral circulation that develops in the presence of an arteriovenous fistula.

A male, age 33, was shot in the left side of the neck and face. After the wound healed, a traumatic arteriovenous aneurysm developed between the external carotid artery and vein behind the descending ramus of the jaw. The aneurysm was of six years' duration. At operation, there was found enormous dilatation of the veins of the neck, with only slight dilatation of the proximal artery. The artery distal to the aneurysm was obliterated. With a temporary ligature around the common carotid, the internal carotid, and the external carotid artery, which cut off all blood supply to the arterial tree on that side of the neck, there occurred no diminution in the size of the veins, and the arteriovenous aneurysm continued to function in a normal manner. This demonstrates the extensive collateral circulation that had developed from the opposite side.

Even though we know that in the majority of cases an extensive and adequate collateral circulation develops in the presence of an arteriovenous aneurysm, one is never justified in operating upon the aneurysm before the collaterals have been tested by Doctor Matas' method. Not infrequently, by

this test, one will find that there is insufficient collateral circulation present; therefore, the collateral circulation must be developed. This development of the collateral circulation is accomplished by blocking the symapthetic nerve supply to the extremity as well as the use of the Matas compressor. Postoperatively, there occur, not infrequently, certain degrees of vasomotor spasm which certainly inhibits the normal circulation of the blood distal to the site of the operation. I have noticed this in two cases of arteriovenous aneurysm. The patient had adequate circulation preoperatively, and after operation, on the second day, there occurred slight bluish discoloration of the toes. This was due entirely to vasomotor spasm produced by the trauma of the operation. Both of these patients had their lumbar sympathetic ganglia blocked by novocain which immediately overcame the spasm of the arterial tree. There was immediate change in color from a bluish-red to a good pink color. Therefore, one should watch closely for the development of postoperative traumatic vasomotor spasm following the repair of an arteriovenous fistula. If this phenomenon does occur, one should inject the lumbar sympathetic ganglia with 1 per cent novocain, and the procedure should be repeated every day or every other day until all tendency to vasomotor spasm has been overcome.

Dr. Edgar L. Gilchrist (San Francisco, Calif.): I appreciate Doctor Mason's reference to the case of arteriovenous fistula of the second portion of the right subclavian which I presented before this Association in 1928. I am happy to report that the patient has remained well. When I first saw him the fistula had been present for seven years, and he had the largest swelling of the entire arm, even involving the hand, that I had ever seen. This extremity was useless. His heart was enlarged. He was kept in bed for several weeks, and his arm elevated, which resulted in a marked reduction of the edema in it, and the bed rest proved beneficial to the heart. I feel that these preoperative measures are most important.

I agree with Doctor Mason that the operation, or at least as much as possible, should be performed under local anesthesia. I do not believe that the importance of performing a subperiosteal resection of a portion of the clavicle can be overemphasized. This affords the operator room and he does not have to work in a hole. The clavicle regenerates readily. Fatal hemorrhages have occurred in patients when the surgeon failed to do this, because he was unable to see and tie off the vessels. I shall never forget Halsted's admonition, and every surgeon operating on an aneurysm should always bear it in mind, when he said: "An operator searching for a bleeding point in a pool of blood, and particularly so when embarrassed in his movements by the adherent walls of an aneurysm within which he is working so disadvantageously, presents a distressing spectacle. I would rather devote an additional hour or more to an operation than be caught for a few moments in such a predicament."

ARTERIOVENOUS ANEURYSM (FEMORAL BELOW PROFUNDA)

CASE REPORT

ISIDORE COHN, B.S., M.D.

AND

BEN R. HENINGER, M.D.

NEW ORLEANS, LA.

THE case which forms the basis of this report was referred to Dr. B. R. Heninger, for supposed decompensation of the heart.

Case Report.—J. D., white, male, age 50, a bridge tender, had noticed shortness of breath and dizziness, particularly after exertion, in 1937. His symptoms became progressively worse and, in January, 1938, he consulted a physician. *Past History:* In 1914, he was shot in the left thigh. Twenty years ago, he was refused life insurance because of a heart murmur. The patient states that, during 1926, he had an attack of palpitation and weakness, at which time he was put to bed for several weeks. Several attacks occurred subsequently.

Physical Examination.—February 8, 1938: The patient was a rather small man, weighing about 139 pounds, apparently comfortable. There is evidence of loss of weight. Pupils are round, equal, react normally. There is a distinct shock of the head with each systole of the heart. Marked pulsations of the vessels of the neck. The thorax on the left side is bulging, but no lagging noted on either side. The apex impulse is easily seen, and is noted to be very diffuse and wavy, occupying the third, fourth, fifth and sixth interspaces, inside and outside the nipple line, at least 13 cm. from the median line in the sixth interspace.

No thrill noted over the precardia. On percussion, the left border of the heart is greatly enlarged. No increase in aortic dulness. Heart sounds are totally irregular; considered to be auricular fibrillation (Fig. 2A). Over the entire precardiac region there is heard a loud, rough, systolic murmur, transmitted in all directions. At the base P^{2nd} is louder than A^{2nd} and possibly accentuated. The radials are not sclerosed. Blood pressure equal in both arms, 130/60. Lungs clear. Abdomen protuberant. Liver just palpable and slightly tender. No evidence of ascites. Circulation time 16 seconds (Decholin). Venous pressure 130 Mm. (saline).

An electrocardiographic tracing was taken at once; this showed a ventricular rate of 80—auricular fibrillation QRS .08, slurred QRS in the first lead and the T inverted in all three leads. S-T 1, 2 and 3 depressed.

Impression of Doctor Heninger: *Definite myocardial disease.*

There were three facts which stood out rather prominently:

(1) A sudden onset of impending heart failure without any apparent reason.
(2) There was an association of auricular fibrillation.
(3) Not being able to account for the cardiovascular symptoms up to this time led to the discovery of the arteriovenous fistula.

At this stage in the examination the patient volunteered the information that he had been wounded by a pistol bullet 24 years previously. Further examination made by Doctor Heninger revealed the following facts:

"The entire left lower extremity was much larger than the right, left thigh in its midportion measured 42.3 cm. while the right measured 37.7 cm., the left calf 32.8 cm.—right calf 26.5 cm.

"The left extremity was perceptibly warmer than the right. On the inner aspect of the upper third of the left thigh, a distinct thrill could be felt.

"The thrill and bruit could be felt and heard in the left side of the abdomen.

"The first roentgenologic examination showed the transverse diameter of the heart to be 18.3 cm. while the hemithoracic diameter was 15.75 cm. (Fig. 1)."

Doctor Cohn saw the case in consultation, February 8, 1938, at which time the following notes were made: "Abdomen: There is an expansile pulsation in the lower left quadrant of the abdomen. Palpation reveals a thrill in the lower left quadrant of the abdomen.

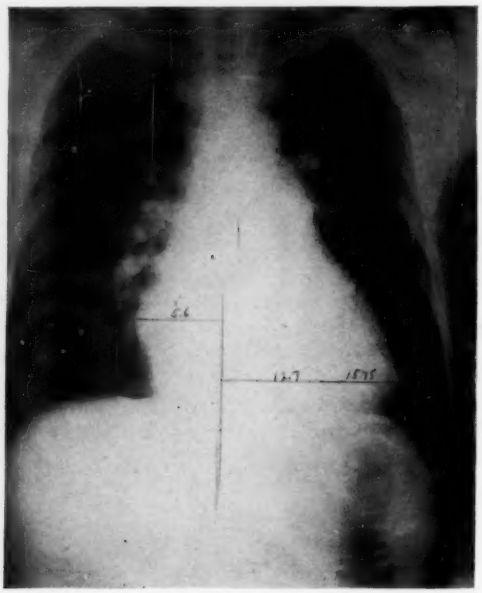


FIG. 1.—Roentgenogram of the chest, February 8, 1938, showing the enlargement of the size of the heart.

"The left lower extremity is larger than the right, and there is a marked pulsation on the inner aspect of the left thigh. On palpation, a definite thrill is felt, and on auscultation, a roaring bruit is heard. The thrill and bruit can be controlled and stilled by pressure. There is also a definite rise in the blood pressure by closing the fistula, and a slowing of the pulse rate (Branham's syndrome)." The measurements of the lower extremity confirmed those which had already been made.

The following impressions were noted at the time:

(1) Patient showed signs of circulatory embarrassment.
(2) A large heart was evident roentgenologically.
(3) There was evidence of auricular fibrillation and definite myocardial disease.
(4) The involved extremity was larger than the normal one.
(5) There was a visible expansile pulsation in Scarpa's triangle.
(6) There were scars of the bullet wounds.
(7) There was a thrill imparted to the palpating hand along the entire length of Scarpa's triangle.
(8) There was a bruit which sounded like the roaring noise made by a steamboat either making a landing or pulling out from shore.
(9) The thrill and roar were stilled by pressure above or over the fistula.

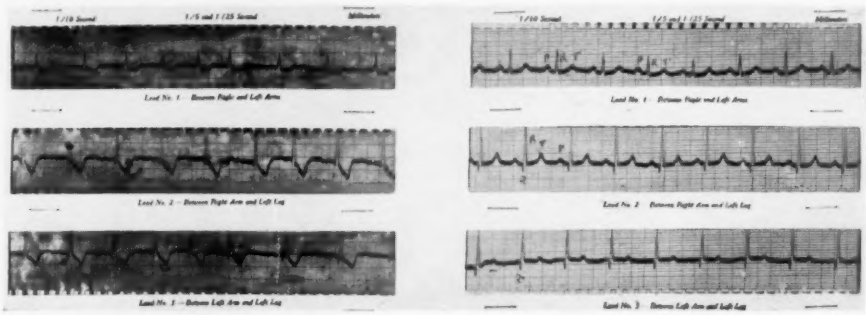
(10) Obliteration of the fistula caused an immediate rise in the systolic blood pressure.

With this evidence before us, the problems of management were clear, if one follows the teaching of Matas.

The following course was adopted:

(1) All available laboratory data were obtained.
(2) Complete investigation of the cardiac disorders by Doctor Heninger.
(3) Complete rest.
(4) Efforts were immediately begun to assure ourselves that collateral circulation was adequate.

Laboratory Data.—February 8, 1938: The total red count was 4,610,000; leukocyte count 6,000; hemoglobin 80 per cent; differential leukocyte count—neutrophils 74 per cent, lymphocytes: small 23, large 0, eosinophils 3 per cent. P.S.P. first hour 60 per cent; second hour 25 per cent. Urine Examination: Straw color, specific gravity



A B

FIG. 2.—(A) Electrocardiogram on admission, February 8, 1939. Note absence of P waves. T wave inversion in all Leads results from administering digitalis.
Note: On February 19, 1938, a sinus rhythm was obtained, after 18 cat units of thevetin and 64 gr. of quinidine sulphate had been administered.
(B) Electrocardiogram showing continuance of sinus rhythm, even though all medications had been discontinued. Note P waves and the upright T waves in all Leads. The digitalis effect has disappeared.

1,020, no albumin, no sugar, negative sediment. Blood chemistry, February 9, 1938: Total nonprotein nitrogen 32.4, dextrose 102, Wassermann negative.

"It was felt from the study of this case that heart failure was inevitable, and that success could only be achieved by correction of the arteriovenous fistula. While the case was being prepared for surgery ample opportunity was provided for trying to establish a sinus rhythm of the heart and build up, if possible, the cardiac reserve, thus making the patient a more favorable surgical risk. Thevetin 4 cc. was given intravenously daily for six days; this total dose is equivalent to 18 cat units; but auricular fibrillation persisted. Quinidine sulphate grains 4, four times daily by mouth, was then given. On the fourth day, a sinus rhythm was obtained, the total dose of quinidine given was 64 grains (Fig. 2 B). After sinus rhythm was obtained, the patient was kept on a maintenance dose of quinidine sulphate grains 8 per day, with relief of all symptoms pertaining to the heart" (Heninger).

In our investigation of the collateral circulation, the Matas compressor was applied daily to the femoral just below Poupart's ligament. Prior to applying the compressor, however, an Ace bandage was applied from the toes up and *it was regularly noted that the blood pressure became markedly elevated even before applying the compressor. After applying the compressor and removing the Ace bandage, the systolic pressure did not remain as high.*

Our impression was that there was an arteriovenous fistula below the origin of the

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FIGS. 3 TO 12.—DRAWINGS SHOWING THE CONSECUTIVE STEPS OF THE OPERATIVE PROCEDURES AND THE OPERATIVE PATHOLOGY FOUND.

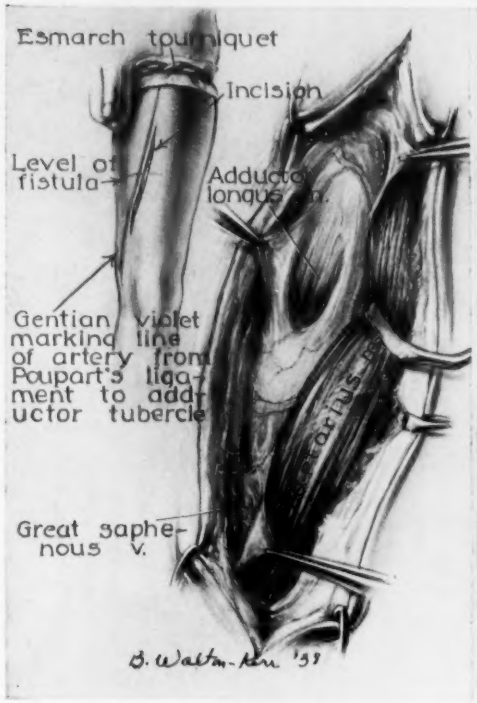

FIG. 3.

FIG. 4.

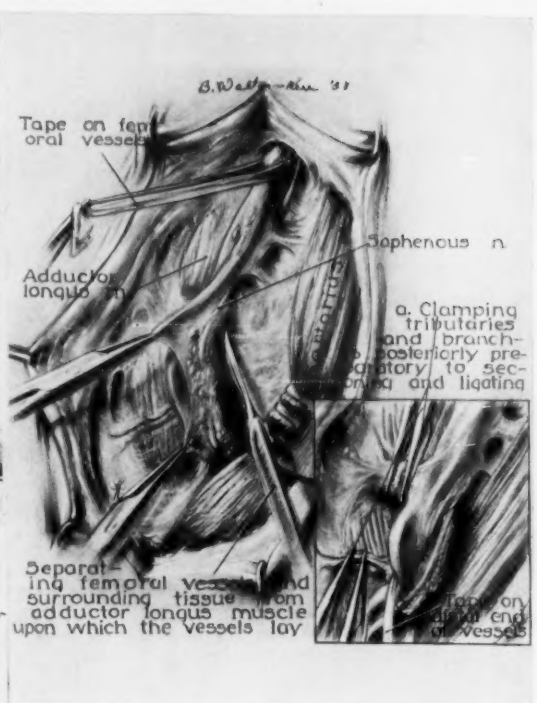

FIG. 5.

profunda. Branham's syndrome was noted from the first day on, *i.e.*, pulse rate dropping after obliteration of the fistula from 80 to 60.

Roentgenogram, March 15, 1938: "Reexamination of the chest shows a considerable decrease in the heart diameter which at this time measures 17.6 cm. The heart diameter, therefore, appears to be slightly decreased as compared with the last examination."

The patient has improved so much with rest in bed and the apparent improvement in circulation was so satisfactory that it was decided to operate. *Preoperative Diagnosis:* Arteriovenous aneurysm, femoral, about the upper limit of Hunter's canal.

Operation.—March 16, 1938: The line of the artery from Poupart's ligament to the adductor tubercle was marked off with gentian violet and the level of what we believed to be the location of the fistula was also marked off with gentian violet by a line transverse to the direction of the vessel. The limb was then elevated and Ace bandages applied from the toes to almost the groin. At this point, the Esmarch tourniquet was applied. The Ace bandages were removed and an incision was made from about two inches below Poupart's ligament to two inches above the internal condyle. The sartorius muscle was identified and dissected free and retracted laterally. At this point, we were able to *identify the vein by the thin wall* and by the bluish color; lying over and superimposed upon the vessel was the superficial femoral nerve. We next identified the roof of Hunter's canal by the direction of the fibers. These fibers were incised. It was noted at this time that neither the vein nor the artery was adherent to the sartorius muscle at this point. After cutting the fibers of the roof of Hunter's canal, we noted a definite bulge of the vein, forming almost a hemispheric dilatation on the medial aspect. Below the level of this dilatation we were able to identify the artery and the vein, and at this stage, a tape was placed around the artery and vein, below the dilatation, and tape was placed around the artery and vein about the middle of Scarpa's triangle, below the level of where we were sure the profunda had been given off.

Having clamped and sectioned between the clamps and ligated small vessels passing in the field, the vessel in its entire length, between the tapes, was separated from the abductor longus muscle upon which it lay. The tributaries and branches posteriorly were sectioned between clamps and ligated. Chromic catgut was used.

At this point, the tourniquet was removed; there was no bleeding. The tissues, in other words, were not stained, but we noticed a huge, fusiform dilatation of the vessel from the level of the tape down to the level of the hemispheric dilatation in Hunter's canal. Below the level, the vessels were not large, in fact, not one-third the size of the upper limit of the vessel. About 20 cc. of thoratrast was injected into the vessel below the level of the upper tape, and a roentgenogram taken.

Two black braided silk ligatures were applied below the tape, about one-third inch apart, and in between this a piece of ribbon catgut was used as a band ligature. The same procedure was adopted below the level of the aneurysmal dilatation—*it should be noted that the ligatures were put on while the tape was held taut, so that a great deal of pressure would not have to be used to approximate the vessel walls.*

We next made an incision into the aneurysm on the venous side between the ligatures. In order to avoid a great deal of bleeding and staining of the tissues, suction was employed to empty the blood in that segment of the artery and vein between the ligatures. In the upper third of the segment, we noted two small openings, and apparently on different levels, one medial and below the other (like a subway at different levels). *The vessels were so fused in this dilatation that it was impossible to separate them and to ligate the artery and vein independently.* Since all bleeding had been controlled, and the arteriovenous aneurysmal segment was freed from the underlying tissues, it was decided to extirpate it, and this was done; there was no bleeding.

Beyond the ligatures, distal to the distal ligature on the proximal end and proximal to the ligatures of the distal end, a transfixion suture of chromic catgut was used, as "hypnotic" ligatures.

The color of the foot was excellent and the dorsalis pedis and posterior tibial pulses

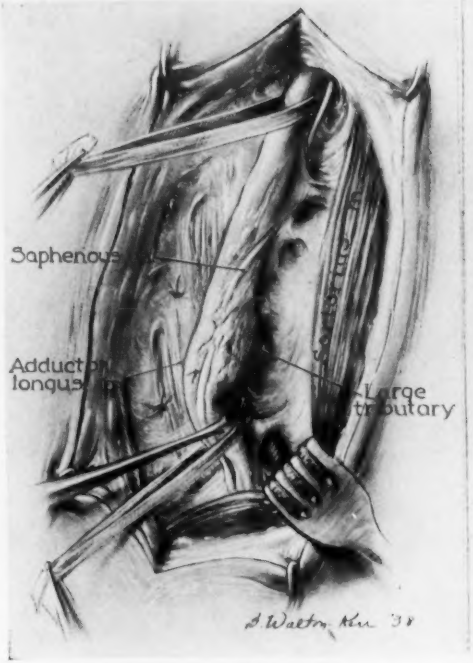

FIG. 6.

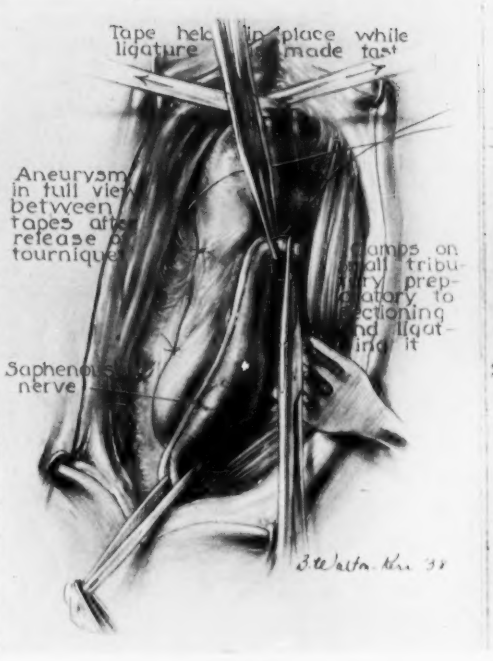

FIG. 7.

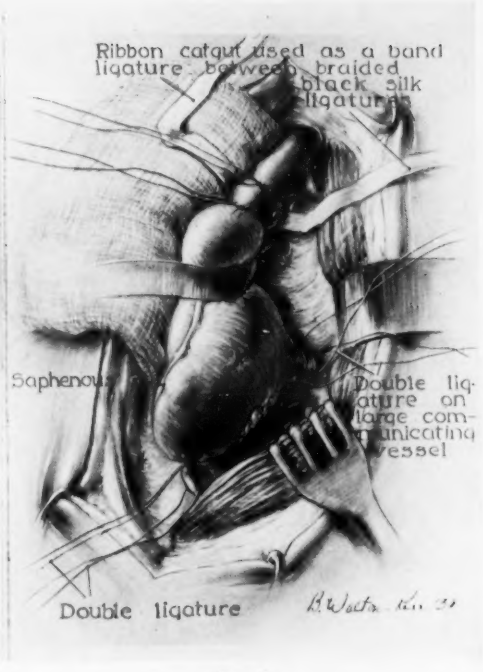

FIG. 8.

were easily palpable. The wound was then closed without drainage; two layers of sutures, plain catgut, interrupted sutures in the fascia, and the skin was closed with interrupted Burdick-Blair-Davis sutures. A copious dressing and a Matas mattress were applied.

The genius of a man is often expressed in a single sentence or paragraph. Such an expression of an individual's attitude is well summarized in the following sentences, taken from the Inaugural Address of Doctor Matas, as President of the American College of Surgeons, in 1926. Speaking of aneurysms he said: "The surgeon should be eclectic in his attitude. He should choose the simplest means by which he may suppress the disease." Again, in 1926, Matas said: "In dealing with arteriovenous aneurysms, the object of the operation will not be accomplished without a complete detachment of the anastomosing vessels or a complete obliteration of the fistula by suture or ligature. To accomplish this purpose modern technic offers many resources, but in a large number of cases the detachment or obliteration of the fistula, with the preservation of the continuity of the artery and vein, is impracticable, and either one or the other vessel, or both vessels will have to be sacrificed."

He further states: "The most conservative procedure is a transvenous suture of the orifice in the artery, with the excision of the detached segment or vein. In many cases the fistula's communication is so buried in a mass of organized exudate of scar tissue that the only alternative left is to do a quadruple ligature, with excision or section of the vessels at the seat of the anastomosis."

These introductory remarks are offered as an explanation of why this case was operated upon by means of multiple ligatures and extirpation rather than by endo-aneurysmorrhaphy. The conditions found at operation seemed to preclude the possibility, at least in my hands, of a successful surgical procedure other than multiple ligation and extirpation of the arteriovenous aneurysm.

Because of the voluminous literature which has developed within recent years on the systemic effects of arteriovenous aneurysm, it would be out of place here to do more than present the evidence of the systemic cardiovascular manifestations presented by this particular case.

The writings of Matas, Reid, Holman, and others on this subject contain all of the known facts; repetition is unnecessary. All that is worthwhile is to present the evidence before us in this case and the result obtained.

During the period that the patient was under observation, the improvement of good collateral circulation indicated that we would be safe in operating.

At operation we found a fusiform dilatation of the artery down to just below the level of the fistula. The vessels seemed so intimately connected that it was considered hazardous to attempt to find any line of cleavage between the artery and vein.

The aneurysmal-like dilatation of the artery above the fistula, the intimate fusion of the artery and vein above and medial to the fistula, plus the large

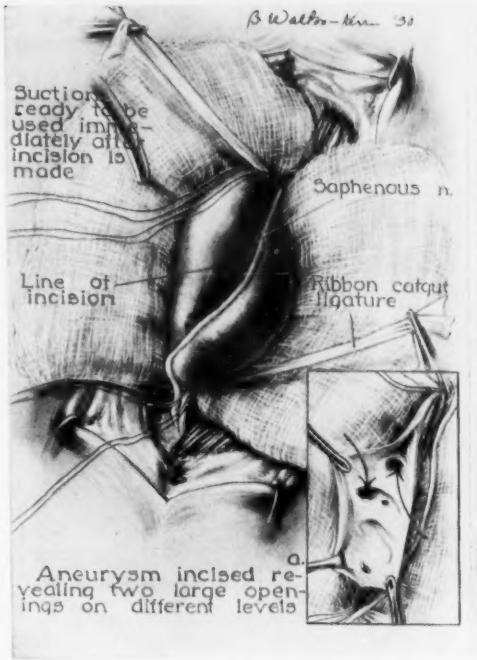

FIG. 9.

FIG. 10.

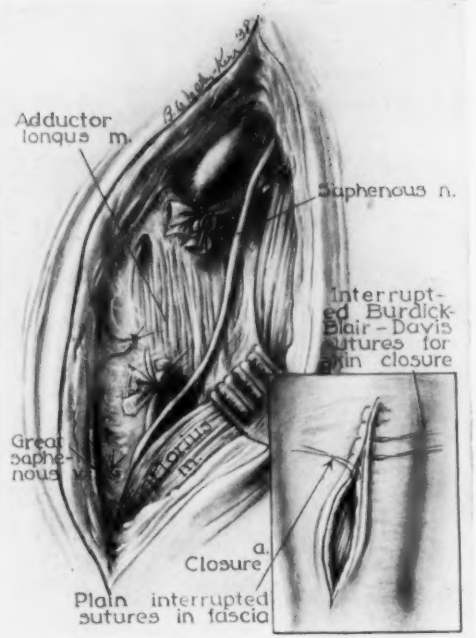

FIG. 11.

size of the fistulous opening and the character of the walls of the fistula, made it imperative, at least for me, to continue with the extirpation rather than to attempt to suture. Subsequent study of the specimen showed that the walls of the fistula contained calcareous deposits. This, I believe, would have added to the difficulties if I had attempted to perform the Matas operation.

Two precautions were observed in the operative management of the case. At the time we were ready to tie the ligatures, the tape above was held taut so as to obliterate the vessel and prevent the pounding of the great volume of blood against the ligatures which would have increased the amount of pressure necessary to ligate the vessel. With the tape on at the time, there was no difficulty in applying the silk ligatures.

FIG. 12.

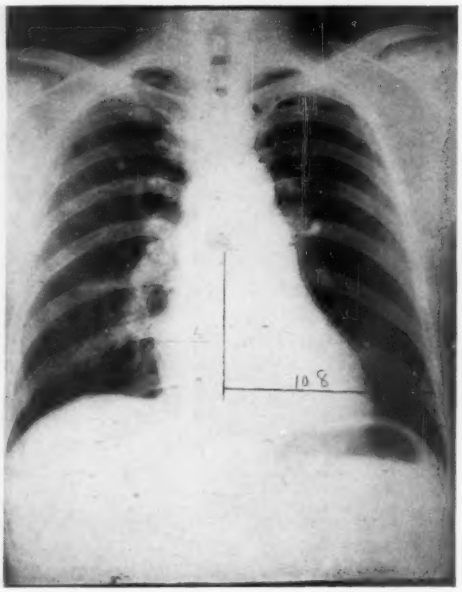

FIG. 13.—Roentgenogram of the chest, July 14, 1938, showing the decrease in the size of the heart.

A second precaution observed was the use of suction at the time that the incision was made in the venous side of the sac; in this way we were enabled to see the interior of the sac with ease and without staining of the surrounding tissues. After the ligatures had been tied on the upper and lower pole of the aneurysm, and all secondary branches and tributaries had been ligated, we found that the sac could be extirpated without difficulty.

We found it advantageous to use a tourniquet, so as to be able to expose the vessel in a dry field. All of the tributaries to the vein could be seen and ligated, and an additional advantage was that the tape could be used to obtain the effect of a tourniquet.

Postoperative Course.—The dorsalis pedis and posterior tibial pulses, immediately following the operation, were easily palpable and remained palpable throughout the subsequent course.

On the second postoperative day, the patient's temperature reached 101° F., and for two weeks he ran a temperature, with morning and evening remissions as high as 102° to 99° F.

On the fourth postoperative day, because of the persistence of temperature, the wound was inspected. At this time we noted that there was some pitting edema of the entire lower extremity. The skin of the leg had a dusky hue, but the sole of the foot had an excellent, almost normal appearance. At this time, the dorsalis pedis and posterior tibial pulses could easily be palpated. Inspetcion of the wound showed that there was no redness surrounding the wound, no discharge from it, no tenderness in the inguinal region, and it was noted that the femoral pulsation below Poupart's ligament did not seem to be as great as it was prior to the operation.

The wound was dressed and a Matas mattress reapplied.

On the tenth postoperative day, the sutures were removed; the wound had healed by primary intention.

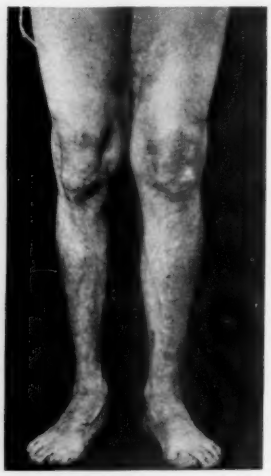

FIG. 14.—Photograph of the lower extremities showing their relative size, six months postoperative.

Because of the persistence of the temperature, laboratory investigations were made to determine, if possible, its cause. Agglutination reactions were done; they were all negative; no evidence of malaria was found and blood cultures, repeatedly done, were negative after 72 hours' incubation. The temperature gradually subsided after about the third week. The edema subsided, and the color of the leg remained good.

Interpretation of the temperature remained a problem, and the only opinion that I have is that it possibly was associated with some thrombophlebitis.

By April 1, 1938, no bruit could be heard over the iliacs, and electrocardiographic tracings at this time showed evidence of the disappearance of the auricular fibrillation. The patient remained in the hospital until May 13, 1938, all the time in bed, or in a chair.

Course Subsequent to Leaving the Hospital.—June 14, 1938: No difference in the length of the two limbs. No difference in the color of the feet. Some glossiness of the leg, none of the thigh. Left thigh is still larger than the right, left leg is slightly larger than the right. On palpation, the dorsalis pedis and posterior tibial pulses are easily felt. On the inner side of the thigh in the region of the original scar, there is some slight thickening on either side of the scar, but the skin moves freely over the underlying muscles. There is no pulsation palpable. The pulsation just below Poupart's ligament is not marked. In fact, it almost feels like the pulsation of a normal femoral artery. There is no thrill imparted to the examining hand. No bruit is heard over the femoral or iliac region. Maximum apex impulse noted in the fifth interspace. Heart sounds are clear, regular, and there are no murmurs. Blood pressure 120/90.

July 14, 1938: Patient has been perfectly comfortable since his last examination, June 14, 1938. He has had no respiratory discomfort or pains in his chest. The only discomfort experienced is a slight tingling sensation just below the left knee.

Examination of the Chest.—I am unable to find any increase in cardiac dulness (Fig. 13), heart sounds seem to be clear and regular, not accentuated and no murmurs. Blood pressure 129/98.

There was no noticeable swelling of the leg, although there is the difference previously noted in the size of the two limbs; measurements show that there is about 2 cm. difference in the circumference of the thighs and relatively the same amount in the calf of both legs. He is able to flex his knee well beyond the right angle and extend it completely (Fig. 14). The dorsalis pedis and posterior tibial are easily palpable and the pulsation

just below the groin in Scarpa's triangle is hardly different from that of the normal femoral pulsation. There is no localized infiltration of his skin about the scar, and no edema. He has gained a great deal of weight.

It should be particularly noted in this examination that I could find no thrills in the femoral region and no bruit over the femoral or iliac artery.

November 8, 1938: Patient has gained 62 pounds. He is working 12 hours a day, and walks without a limp. The heart sounds are clear and no murmurs are noted. There is slight difference in the size of the two lower extremities. Dorsalis pedis and posterior tibial pulses easily palpable. Blood pressure 128/80.

Summary of Medical Points of Interest (B. R. H.)

(1) Size of heart, murmurs and lack of background for factor in producing heart disease lead to discovery of an arteriovenous fistula.

(2) The association of auricular fibrillation with an arteriovenous fistula is extremely rare.

(3) Why the sudden onset of symptoms pertaining to the heart, in the face of an arteriovenous fistula of 24 years' standing?

(4) If arteriosclerosis is the cause of a heart disability, why has the heart appeared adequate, for so long a time, in spite of the arteriovenous fistula and cardiac enlargement?

(5) Definite Branham's syndrome, after restoration of sinus rhythm.

(6) Reversibility of the heart as to size, after correction of the arteriovenous fistula by surgery.

(7) Very little evidence of heart disease, including cardiogram and exercise tests, following recovery from surgical correction of arteriovenous fistula.

SUMMARY

(1) An arteriovenous fistula was present for many years, which did not incapacitate the patient until a few months before he came under observation.

(2) The presence of the fistula was not recognized prior to his admission to the hospital.

(3) After many years, there was evidence of circulatory embarrassment, associated with an enlarged heart, auricular fibrillation, and definite electrocardiographic changes.

(4) An excellent collateral circulation had developed.

(5) Following rest in bed and medical care, the size of the heart diminished and the patient's general condition improved. Branham's syndrome was noted throughout the preoperative period.

(6) Marked rise of systolic and diastolic pressure followed obliteration of the fistula.

(7) Following the teachings of Matas, a period of rest in bed, and study of the efficiency of the collateral circulation was made before operation was performed.

(8) The tourniquet was used in order to obtain a dry field.

(9) Because of the fusion of the two vessels and because of the rigid walls

of the fistula, it was deemed wise to perform multiple ligations and extirpation, rather than to attempt to perform an endo-aneurysmorrhaphy.

(10) Following operation, the circulation in the limb was in no way impaired.

(11) The condition of the heart has improved tremendously so far as size, rate, and rhythm are concerned. Electrocardiographic changes now do not indicate evidence of myocardial damage.

(12) The patient has returned to his normal activities.

Discussion.—Dr. R. H. Jackson (Madison, Wis.): I am asking for information and counsel. I think the surgeon who is faced with performing an operation for arteriovenous aneurysm of the femoral vessels for the first time, does so, if he reads the literature, with a good deal of apprehension. The literature is full of warnings, and he is advised not to put any other operation on that morning's list as it may take several hours and should be performed under local anesthesia. The danger of catastrophic hemorrhagic complications is reiterated.

The method which, in my very limited experience, has proved most efficacious in obviating the danger of (1) complicating hemorrhage; (2) the tediousness of a prolonged operating period; and (3) the nerve-wracking "peckiness" associated with performing such an operation under local anesthesia, is as follows:

(1) An Esmarch bandage is applied from foot to groin, Wyeth hip amputation pins are inserted and a tourniquet placed above them and the Esmarch removed. This insures a dry field for dissection.

(2) Full exposure of the arteriovenous aneurysm is quickly accomplished (Fig. 1); multiple ligatures are applied (Fig. 2); and the lesion excised (Fig. 3).

(3) The tourniquet is removed and the wound closed.

Does this method carry with it any danger that the application of the Esmarch bandage will throw enough additional blood into the circulation to cause a sudden dilatation of an already greatly enlarged and embarrassed heart? There was on evidence of this.

Finally, with all due respect to Doctor Royster and the majority of the Fellows who, two years ago, turned "thumbs down" on the use of spinal anesthesia, I advocate the administration of this method of anesthesia as an ideal one in this particular field.

Dr. J. E. J. King (New York, N. Y.): In Germany, in 1915–1916–1917, we had nine cases of aneurysm. Four of these were arteriovenous aneurysms involving the femoral vessels; they were low in the apex of Scarpa's triangle and were below the origin of the profunda. They were practically all alike except that in one a dumb-bell-shaped piece of a bullet jacket was lodged in such a manner that it would not go in either direction. It had been polished smooth by the blood stream and was bright and shiny. The femoral nerve was not injured in any of the four cases. Quadruple ligation was performed in all, with excision of the intervening portions of artery and vein, including the fistulous tract. All recovered without loss of foot or toes and walked well.

At that time, we did not know enough to close the fistulous opening in the artery through the vein by sacrificing the vein and preserving the artery, or any other method. In addition, there was a case of subclavian aneurysm in which a flap including a portion of the clavicle was made in order to expose the sac. This was readily accomplished, and soft clamps were applied proximal and distal to the sac, but when the sac was opened for the purpose of evacuat-

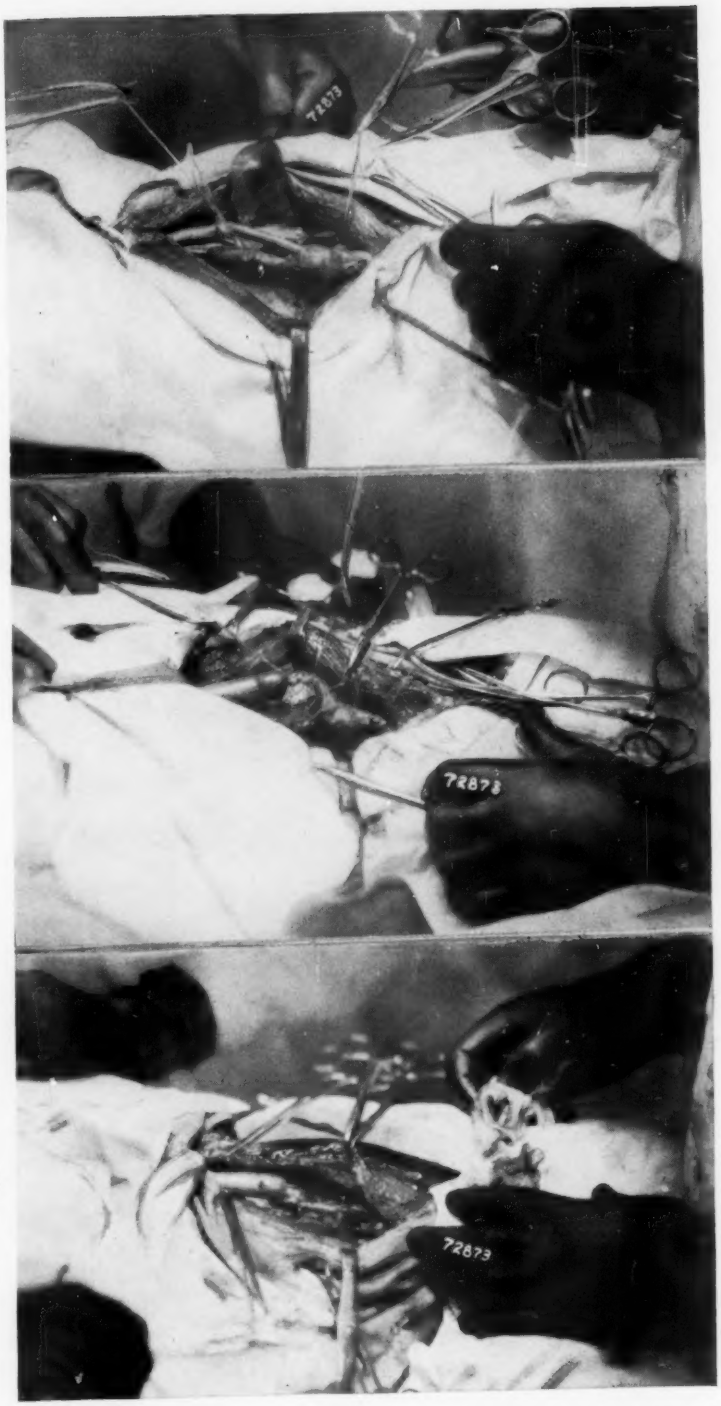

FIG. 1.—The arteriovenous aneurysm is quickly exposed in a dry field. (2) Multiple ligatures are applied and the tourniquet released. (3) The arteriovenous aneurysm is excised.

ing clots and repairing it, there was profuse bleeding from numerous small vessels, the openings of which "pepper-boxed" the floor of the sac. It was impossible to control bleeding, and at that time transfusion was not readily performed. The patient died of loss of blood and shock. Proximal ligation probably should have been undertaken.

DR. JOHN W. PRICE, JR. (Louisville, Ky.) : I became interested in vascular surgery shortly after Carrel reported his method of doing end-to-end vascular suture. We perfected our technic after Crile's method had been tried and made some further modifications by use of a cannula similar to that of Crile, which could be put on a vessel; the vein was cuffed over the cannula, the artery pulled over the cuff and sutured to the vein; later the cannula was removed. We performed many anastomoses between the carotid vessels of the neck. We began doing this in 1908 and continued it for four or five years.

It was not until after the War that I had my first case of arteriovenous aneurysm to operate upon. I had gone through the War and operated upon every sort of gunshot wound and found nothing suitable. Finally a young fellow was brought in, shot below Poupart's ligament. I thought if only a small hematoma developed in the thigh I had a case suitable for arterial suture. I put him in the hospital and waited about three weeks, which was a reasonable time to watch for the occurrence of any infection which might have been carried into the wound. He went along nicely with no elevation of temperature, and we made an exposure and found an enormous arteriovenous aneurysm, which seemed to me to be unsuitable for suture. We performed multiple ligation of the femoral artery and vein and excised the whole mass. He returned to work very shortly afterward; that was 15 or 16 years ago, and he is still working.

SUPPURATIVE PERICARDITIS

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A NUMBER of excellent papers on suppurative pericarditis have appeared in the American literature during the past few years, among them those presented before the Southern Surgical Association by Dr. George H. Bunch,[1] in 1934, and Dr. Arthur M. Shipley,[2] in 1935. Most of the authors have stressed the importance of early surgical drainage and have indicated that it is the only method of treatment which gives the patient with pus in the pericardial sac a reasonable chance of recovery. In spite of this, it appears that drainage of suppurative pericarditis is still an unusual event in most hospitals.

No doubt the explanation is that pericarditis is difficult to diagnose. That it is not especially rare is shown by a study of autopsy statistics. Pyrah and Pain[3] found 214 cases of acute pericarditis among 7,965 autopsies performed at the Leeds General Infirmary between 1921 and 1931. Ninety-one of these, or more than 1 per cent of the total number of autopsies, showed acute suppurative pericarditis. That pericarditis is difficult to diagnose is also shown by autopsy statistics. Cabot[4] has stated that 77 per cent of all cases of pericarditis found at autopsy at the Massachusetts General Hospital were not diagnosed before death.

The records of the Medical College of Virginia Hospitals show that the diagnosis of acute pericarditis has been made in 50 cases during the past eight years. Twenty-six of these were made by the clinicians and 24 were first made by the pathologists. In other words, it appears that 50 per cent of the patients with acute pericarditis in our hospitals are not diagnosed clinically, this in spite of the fact that members of both the medical and surgical staffs are especially interested in diseases and injuries of the heart and pericardium.

Of these 50 cases of acute pericarditis, 17 were either acute fibrinopurulent or purulent. Seven of the frankly purulent ones were correctly diagnosed and six were drained. In one case the cartilages were resected preparatory to drainage, but it was found that the pleura extended over beneath the left border of the sternum and in attempting to dissect it away from the pericardium the pleura was accidentally opened. Unfortunately, it was decided to pack the wound until the following morning, to give the two layers of pleura an opportunity to adhere. They were then to be sutured and the pericardium opened. To our chagrin, the child died during the night.

Only four of the ten cases first diagnosed at the autopsy table had a demonstrable increase in intrapericardial fluid; death in the remaining six occurred from an overwhelming infection before there had been an opportunity for pus to form in the pericardial sac. All four of the patients who

were found at autopsy to have a demonstrable increase in pericardial fluid were moribund when admitted to the hospital; three of them died within less than 12 hours thereafter, and the other lived only 24 hours.

One of the chief factors in the failure to diagnose acute pericarditis is the absence of significant symptoms. In suppurative pericarditis, which is almost always secondary to infection elsewhere, the picture is likely to be obscured by the associated or antecedent disease. This is especially true when the primary infection is within the thorax, as is so often the case.

It is obvious, therefore, that the correct diagnosis will not often be reached unless physicians make repeated examinations of the cardiac area in septic cases, especially those with intrathoracic or subphrenic infections, and in osteomyelitis with septicemia.

The physical signs are usually more characteristic than are the symptoms. It is probable that a friction rub occurs at some time in all such cases, and it not infrequently persists even in the presence of a large accumulation of fluid; but in other instances it is transient. If a friction rub is not heard and there is no appreciable increase in the amount of intrapericardial fluid, the diagnosis cannot be made, but fortunately, in those cases where the diagnosis is of the greatest importance, there is a rapid increase in the amount of fluid, which produces the signs of cardiac tamponade. In such cases, physical examination will show an increase in the area of pericardial dulness and roentgenograms demonstrate an enlarged pericardial shadow, while fluoroscopic examination will show an apparent immobility of this shadow. These signs are strong evidence of the presence of a pericardial effusion, but that an effusion is purulent can be proved only by pericardicentesis or pericardiostomy. The following case is illustrative:

Case Report.—M. W., white, male, age seven, entered the Memorial Hospital, March 4, 1938, with the signs of a massive empyema of the right pleural cavity. He also had marked weakness of the right arm; edema of the right chest wall; and enlargement of the liver. He was having great respiratory difficulty; aspiration was performed immediately and 500 cc. of pneumococcic pus removed. Aspiration was again performed the following morning and 300 cc. of pus removed, and that afternoon the right pleural cavity was drained. The liver decreased in size for several days and then began to enlarge again. On the ninth day after operation it was noticed that the heart sounds were distant and a teleoroentgenogram showed marked enlargement of the cardiac shadow. That afternoon the pericardium was aspirated and 200 cc. of hemorrhagic fluid removed. Following removal of the fluid the symptoms of tamponade promptly disappeared, but continued to recur, and during the next five days a total of 870 cc. of the same character of fluid was aspirated. Repeated cultures were consistently negative. The liver continued to enlarge and ascites developed. Death occurred, April 28, 1938, and autopsy showed an empyema cavity, with no walled-off pus; complete obliteration of the pericardial space by fibrous adhesions; gangrenous perforation of the right diaphragm; extensive fatty degeneration of the liver; splenic tumor, and ulcerative colitis.

The clinical picture in this case was strongly suggestive of suppurative pericarditis but the repeatedly negative smears and cultures proved that the fluid was noninfectious. The cause of the hemorrhagic effusion was not determined.

Because of the difficulty of making a positive diagnosis of intrapericardial suppuration, we have done a diagnostic aspiration in five of the nine cases we have drained; but if the diagnosis seems clear, it is probably better to proceed with the pericardiostomy without preliminary puncture, as pericardicentesis is not without danger.

Shipley[5] and others have shown that approximately 50 per cent of the patients with suppurative pericarditis who are given adequate drainage will recover, but, even though most of those who die following drainage die from associated infections rather than from the pericarditis, there seems to be a hesitancy on the part of many physicians in referring patients for pericardiostomy.

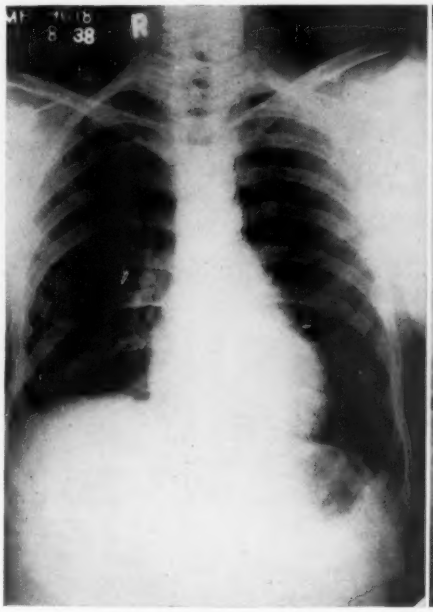

FIG. 1.—Case 8, Table I, C. W.: Teleoroentgenogram, two years and eight months after operation, showing a normal cardiac shadow.

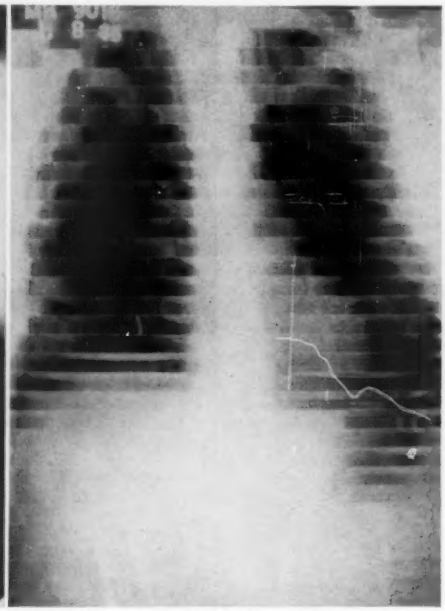

FIG. 2.—Case 8, Table I, C. W.: Roentgenkymogram, two years and eight months after operation, showing almost normal cardiac mobility.

With this in mind, it seems unfortunate that there have been several reports in the recent literature of patients with suppurative pericarditis who have recovered following aspiration alone or in combination with the administration of sulfanilamide. It does not seem reasonable that an appreciable percentage of these patients will recover without drainage, as there is no apparent reason why suppurative pericarditis should respond to aspiration any more satisfactorily than suppurative pleurisy; and, while an occasional patient with pleural empyema recovers following aspiration, it is the exception and not the rule. In early infection of the pericardium by hemolytic Streptococci, aspiration plus the administration of large doses of sulfanilamide may result in a larger number of cures than in infections by other organisms; but even in

TABLE I

EIGHT CASES OF SUPPURATIVE PERICARDITIS TREATED SURGICALLY*

Patient	Antecedent Disease	Infecting Organism	Known Duration of Pericarditis	Type of Operation†	Drainage Material	Method Used to Prevent Pocketing of Pus Posterior to Heart	Immediate Result or Cause of Death	Late Result
L. Y., white, female, age 10. (Univ. of Va. Hosp.)	Pneumonia. Bilateral empyema	Hemolytic Streptococcus	1 mo.	Resection of fifth cartilage. Suture of pericardial edges to fascia	None	Daily passage of lubricated catheter around heart	Recovery	No symptoms referable to heart 12 yrs. later
D. H., white, female, age 5. (Vanderbilt Univ. Hosp.)	Osteomyelitis of femur. Septicemia	*Staphylococcus aureus*	2 da.	Resection of fifth and sixth cartilages. Suture of pericardial edges to fascia	None	Daily passage of lubricated catheter around heart	Recovery	Unknown
G. McD., white, male, age 2. (Med. Coll. of Va. Hosps.)	Osteomyelitis. Septicemia	*Staphylococcus aureus*	8 da.	Resection of fourth and fifth cartilages. Suture of pericardial edges to fascia	None	Daily passage of lubricated catheter around heart	Death 16 da. after pericardiotomy. Septic infarcts in both lungs; peritonitis; endocarditis	
A. McD., white, male, age 14. (M. C. Va. Hosps.)	Carcinoma of left main bronchus. Empyema	*Staphylococcus aureus*	14 da.	Resection of fifth and sixth cartilages. Suture of pericardial edges to fascia	None	Daily passage of lubricated catheter around heart	Death 21 da. after pericardiotomy. Obstruction of inferior cava. Septic infarcts in lung	
J. K., white, male, age 12. (M. C. Va. Hosps.)	Suppurative arthritis. Septicemia	*Staphylococcus aureus*	24 hrs.	Resection of fourth and fifth cartilages. Suture of pericardial edges to fascia	None	Digital separation of fibrinous adhesions	Death 3 da. later. Septicemia	

E. S. M., white, female, age 10. (St. Luke's Hosp., Richmond, Va.)	Pneumonia. Empyema, right	Nonhemolytic Streptococcus	? Pericardiostomy on day of admission	Resection of fourth and fifth cartilages. Suture of pericardial edges to fascia	None	Daily digital separation of fibrinous adsions	Recovery	No cardiac symptoms or signs
J. D., colored, male, age 16. (M. C. Va. Hosps.)	Tuberculous pericarditis. Secondary Streptococcic infection	Nonhemolytic Streptococcus	About 1 mo.	Resection of fifth and sixth cartilages. Suture of pericardial edges to fascia	Penrose drains passed on either side	Digital separation of adhesions. Passage of rubber catheter	Greatly improved for about 2 mos. Developed obstruction of inferior cava, which was released. Death as result of abscesses in kidneys; probably septicemia	
C. W., white, male, age 16. (M. C. Va. Hosps.)	Lobar pneumonia. Empyema	Pneumococcus	24 hrs.	Resection of fourth and fifth cartilages. Suture of pericardial edges to fascia	None	Digital separation of adhesions around heart	Recovery. Convalescence prolonged by infection of costal cartilages	No symptoms referable to heart, nearly 3 yrs. later. Working as mechanic

* A more complete report of these cases will be found in a previous publication (South. Med. Jour., **30**: 164, 1937).

† All of these patients were operated upon under local anesthesia.

the Streptococcic cases, it would seem more rational to establish continuous drainage and also administer sulfanilamide. This is especially true since pericardiostomy is a relatively simple operation which can be performed under local anesthesia with little or no shock. It has been stated that exposure of the heart to atmospheric pressure interferes with its function, but Blalock[6] has shown that this is not true. Furthermore, anyone who has watched pus gush out when the pericardium is opened, and has observed the immediate improvement in the circulation, would find it difficult to believe that the open drainage of such cases was contraindicated.

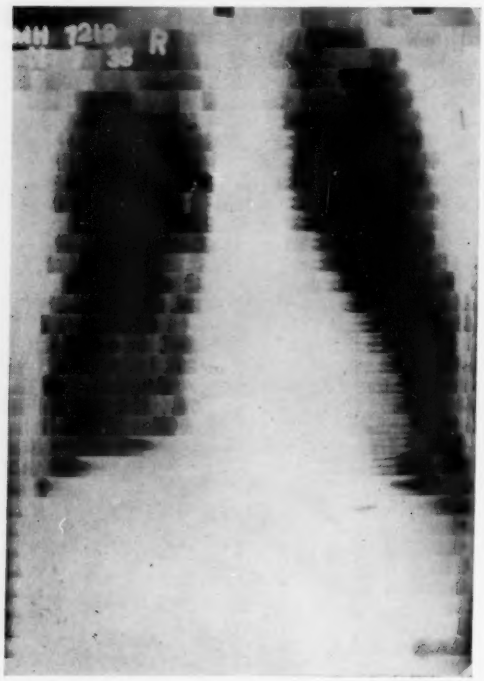

FIG. 3.—Case 9, R. J.: Roentgenkymogram, eight months after operation, showing moderate reduction in mobility of right side of heart.

Pericardiostomy may be accomplished through an intercostal incision, through a trephined opening in the sternum, by resection of one or more costal cartilages either on the right or left side, or by a subcostal, transdiaphragmatic approach. Most surgeons prefer a left parasternal approach, but Lilienthal[7] has suggested a right-sided approach when there is an associated empyema of the right pleural cavity, to avoid the danger of infecting the left pleura. Truesdale[8] and Moore[9] recommend a left axillary approach in order to establish drainage in that portion of the pericardial sac posterior to the heart. This has much to recommend it and should be employed if there is fusion of the two layers of pleura, but is obviously unsuitable in a large percentage of cases because of the danger of infecting the left pleura. Resection of the lower costal cartilages on the left may give more dependent and, therefore, more ideal drainage, but it does not give as ready access to all parts of the pericardial cavity as resection of the fourth and fifth cartilages, and it also carries the danger of infection of the costal arch, which is a rather serious complication.

With these facts in mind, I have usually resected the fourth and fifth left costal cartilages, and have, at times, rongeured away that portion of the left border of the sternum between the third and sixth cartilages. The pleura is carefully freed-up and retracted to the left, to expose the pericardium, which is incised for one and one-half or two inches. The edges of the pericardial incision are sutured to the muscle sheath or the fascia to prevent too rapid

closure of the drainage tract. This gives an excellent exposure of all parts of the pericardial sac and permits digital separation of the fibrinous adhesions between the pericardium and epicardium which form so rapidly in the early cases. One of the chief advantages of establishing drainage before the fluid becomes frankly purulent is the rapidity with which the pericardial sac contracts down and thus obliterates the infected cavity; but to avoid the danger of sacculation of pus posterior to the heart during this time, the pericardium and epicardium are separated at least once in each 24 hours for several days.

I have used this method of drainage in nine patients with suppurative pericarditis. Five have recovered and four have died. In none of the fatal cases was there sacculation of pus posterior to the heart.

These nine cases are presented. Eight of them were reported in a previous publication[11] and are recorded in Table I. One new case is added.

Case Report.—R. J., white, male, age 28, entered the Memorial Hospital, March 1, 1938, with pneumonia of the right middle and lower lobes. Roentgenograms, March 6, 1938, showed fluid in the right pleural cavity, with displacement of the mediastinal structures to the left, and enlargement of the cardiac shadow. On this same day, a pericardial friction rub was discovered. His venous pressure was not appreciably increased at this time but the following day was 15 cm. of water. On March 9, the friction rub was still present but fluoroscopic examination showed a large pericardial effusion. The pericardium was aspirated and 200 cc. of thin pus was removed which showed pneumococci, Type VIII, on smears and cultures; but on March 11, the venous pressure had risen to 19 cm. of water, so immediate operation was advised. Under local anesthesia the fourth and fifth cartilages were resected and the pericardium exposed. When it was opened, between 800 and 1,000 cc. of fluid was removed by suction. The edges of the pericardial wound were sutured to the fascia and no drainage material was used. The adhesions were separated daily for a period of about two weeks by a well-lubricated, sterile gloved finger. The patient made a very satisfactory recovery and was discharged from the hospital, May 6, 1938. He is now working in a tobacco factory and is without cardiac symptoms.

It is impossible to draw conclusions either from the study of a few cases or from the literature, as to the effect of early operation on the prognosis in suppurative pericarditis. This is not surprising, as many factors influence the chances of recovery—the antecedent disease, type of infecting organism, duration of the infection, adequacy of treatment, *etc.* One important point which should not be overlooked is the fact that those patients with the most virulent infections will die without operation if a policy of delay is pursued. This will naturally tend to improve the results if only those operated upon are reported; but, since there is no evident contraindication to early operation as in acute empyema of the pleura, it seems reasonable to suppose that the total number of cures in patients with acute suppurative pericarditis will be greater if they are operated upon as soon as the diagnosis is established. It seems quite clear that the recovery period is reduced by early operation, and it may be significant that the pericarditis was of relatively long duration in our two patients who developed obstruction of the inferior vena cava.

Four of our five surviving cases have been followed from a few months

to 12 years. They are all able to take part in ordinary activities and none of them has any evidence of circulatory embarrassment due to pericardial adhesions. This corresponds with the results reported by Shipley[10] in his own cases and also in those cases which he collected from the literature.

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Discussion.—Dr. Alfred Blalock (Nashville, Tenn.): The most important point regarding the treatment of acute suppurative pericarditis is the value of early adequate drainage. If one waits until the pericardium becomes enormously large and thick-walled, one is more apt to get constrictive pericarditis. As Doctor Bigger indicated, there is no very satisfactory method of draining the pericardium, and it seems to me that taking out the fourth and fifth ribs and part of the sternum will probably result in fewer instances of constrictive pericarditis.

We have had in the Vanderbilt Hospital, during the last 11 years, 33 patients in whom a diagnosis of acute suppurative pericarditis has been made or confirmed at the time of operation or autopsy. The predominating organisms were Staphylococcus, Streptococcus and pneumococcus; rarer organisms have been the Leptothrix, the meningococcus and the diphtheria bacillus. One patient with a Staphylococcus infection, a boy, age 14, with osteomyelitis and septicemia, developed constrictive pericarditis several months following drainage of the pericardium. I thought this to be the first instance of constriction following drainage, but I learned from Doctor Bigger that he had had two similar experiences. We have had another patient who developed signs of increased venous pressure, but this disappeared and removal of the pericardium was not necessary.

It seems to me that Doctor Bigger's point of adequate drainage with removal of two or more costal cartilages will cut down on the incidence of constrictive pericarditis following suppurative pericarditis.

TREATMENT OF PEPTIC ULCER

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IT MUST be admitted that there is no very good reason why the treatment of ulcers of both stomach and duodenum should be classified together; since the treatment in most cases is handled on a somewhat different basis. Likewise, the possibilities of associated cancer with the one and its almost total absence with the other.

The problem of treatment of duodenal ulcer is still unsolved. No method of treatment has been advanced that has uniformly controlled gastric acidity in every case. We continue to have recurrences after all methods of treatment. This would indicate that the acidity has either not been controlled satisfactorily or that the patient is constitutionally predisposed to ulcer. Unfortunately, there is no way to measure tissue resistance to gastric secretion.

In conformity to the generally accepted ideas of treatment, only a small percentage of our cases of peptic ulcer have undergone surgical operation. The great majority have been treated by medical means.

Etiology.—Many causes for the formation of peptic ulcers have been suggested, such as the stress and strain of modern life (Crile's kinetic drive), constitutional predisposition, constitutional susceptibility of tissues to ulceration, trauma of coarse foods, irritative influence of gastric secretions, *etc.* To those above should probably be added, as a cause, gastro-intestinal allergy to certain articles of food which may cause marked irritation in some individuals. If the gastro-intestinal tract is being constantly irritated by these particular foods, it would seem likely that ulceration could result. The fact that peptic ulcers are not by any means unknown in children nullifies to some extent the theory of mental strain and kinetic drive.

Pathology.—While the peptic ulcers seen in this country are almost always single and not as a rule associated with diffuse gastritis, apparently in Europe, multiple ulcers involving both stomach and duodenum, associated with generalized gastritis, are common. In those cases, the edema and thickening of the gastric wall presents a truly remarkable appearance. In two such cases that I have operated upon, the gastric wall in places was from one-quarter to one-half inch in thickness.

Physiology of Gastric Secretion.—It is thought by many, although not as yet definitely proven by laboratory procedures or by experimental animal surgery, as stated by Lewis[1] and others, that the pylorus and the antral end of the stomach, or mucosa therein, exercise control over the acid output of the gastric juice. It is thought, therefore, that resection of the pylorus and antral end of the stomach should definitely cut down acid production.

After operations such as pyloroplasty, gastroduodenostomy, gastro-enterostomy and resection associated with jejunostomy, the lowering of the stomach

acidity is thought to be due in a great part to a reflux from the duodenum bringing about a certain neutralization of the gastric acidity. The amount of reflux and neutralization will obviously be much greater when resection and end-to-side anastomosis with the jejunum has been performed.

Obviously, any operative procedure in the stomach which brings about a radical change in the mechanics of digestion naturally disturbs the normal physiology of the stomach to a considerable extent, and may, for the time at least, be more or less disturbing to the patient. After many such operations, at least a mild form of gastritis probably takes place.

In the course of time the patient's stomach becomes adjusted to the changed conditions and ultimately comfort is usually attained. Postoperative care as to diet and general hygiene is of just as much importance here as in the cases that are depending entirely on medical treatment.

In view of the fact that there is a more or less constant flow from the stomach into the duodenum, probably comparatively little increased reflux of duodenal fluid takes place into the stomach after plastic operations on the pylorus and duodenum.

Indications for Surgery.—For some time, it has been rather generally conceded that in case an ulcer of the stomach fails to heal under adequate medical treatment, it should be treated by surgical methods, such as excision plus gastro-enterostomy or subtotal gastric resection, usually associated with gastro-jejunostomy. Among the reasons for this attitude is, no doubt, the fact that at times, at least, the persistent ulcer proves to be on a cancerous base and, therefore, it is a very serious matter to dally with such a situation, as well as manifestly unfair to the patient. Certainly in the case of suspected malignancy, impending perforation or a type of ulcer that is definitely incurable by medical means, surgery should be resorted to.

Duodenal ulcers in particular appear at times to have seasonal recurrences and recessions. In that location, neither bleeding nor perforation usually takes place until after the ulcer has been in existence a rather long time. Likewise, it is very well established that duodenal ulcers are definitely more amenable to medical treatment than are gastric ulcers.

The choice of surgical operation is a much mooted and discussed point. Many American surgeons apparently favor conservative procedures, more particularly in the treatment of most duodenal ulcers. While another group, also many of the Continental surgeons, apparently favor gastric resection as a method of treatment for the majority of duodenal ulcer cases coming to operation.

When we consider the subject of surgical treatment of gastric ulcer, the location of the ulcer with reference to the greater or lesser curvature has some bearing on the method to be employed. An ulcer situated on the lesser curvature cannot well be locally excised without actually deforming the stomach to a very considerable extent; whereas, an ulcer on the greater curvature can usually be readily excised. This procedure can be accompanied by gastro-enterostomy for purposes of drainage; also for neutralization of gastric acidity.

The large callous ulcer, likewise the deep perforative ulcer, are both probably best dealt with by resection.

If the patient having a painful, chronic duodenal ulcer is not so situated economically that he is able to carry out a proper regimen of dietetic and medical treatment, operation is probably often advisable in such a case.

Chronic, recurring bleeding is a definite indication for some form of radical surgery provided, of course, that the patient's condition is sufficiently favorable to warrant such a procedure. Insofar as the usual routine treatment for chronic, recurring bleeding is concerned, no one has yet been able to approach the favorable results obtained by Meulengracht.[2] In 251 consecutive cases of bleeding from peptic ulcer, there was a mortality of only about 1 per cent, which he contrasted with 289 cases from the same country and the same general class of people treated by the old starvation method; the mortality of the latter group was 7.5 per cent.

Cases of definite stenosis of the pyloric opening are favorable for pyloroplasty or gastroduodenostomy. However, the latter procedure is open to the objection that a resection later on is made more difficult.

Choice of Operations.—In the case of duodenal ulcer associated with a relatively low gastric acidity, it is generally felt that gastro-enterostomy will usually bring about a cure. Likewise, many of these cases are relieved by pyloroplasty of the Horsley, Judd or Finney type, the latter being not, strictly speaking, a pyloroplasty but rather a gastroduodenostomy. In cases in which the total acid content of the stomach is high, the duodenal ulcer constant, and not responsive to medical treatment, gastric resection may be indicated; and if so, about the junction of the middle with the proximal third of the stomach for the point of incision is probably the choice of most surgeons at this time.

Walters[3] has stated that more cases of ulcer will be cured or relieved of their symptoms by gastro-enterostomy than by any other operative procedure. However, if this operation is performed in the presence of high acidity, there is considerable danger of jejunal ulcer developing.

Connell[4] has suggested that those surgeons who consider pyloroplasties and gastro-enterostomies too conservative, and gastric resection too radical, might well resort to fundusectomy in order to cut down the output of gastric acid. So far this method has apparently not had an extensive tryout.

An occasional case of deep perforating ulcer of the duodenum is so surrounded by dense inflammatory tissue that it cannot be properly excised and closed. In such a case I[5] have, on one occasion, tamponed the beginning ulcer sinus with omentum after which a gastro-enterostomy was performed. The final result was satisfactory. Walters[6] has reported a similar case.

The surgery of acute perforations seems to be pretty well standardized in this country. Practically everyone is practicing reinforcement of the suture with an omental patch after closing the opening. This last bit of technic is of great importance, for the reason that a recently repaired perforation may break down or even a second perforation may take place alongside the first. Formerly, a gastro-enterostomy was advised by some as an additional procedure

at this time. This has been quite generally abandoned. However, in cases in which there has been but slight leakage, and that but for a short length of time, and definite pyloric obstruction is present, pyloroplasty or gastro-enterostomy may be indicated at the time. It is also believed, I think, by most surgeons that a perforated duodenal ulcer, after repair and healing, does not usually recur. Judin[7] has reported a large series of perforated duodenal ulcer cases which were treated not only by repair of the perforation but by partial resection of the stomach as well. This would seem to be extremely radical and an unnecessary attack on the patient at the time of such a catastrophe. His reported mortality for all of his cases has been about 12 per cent.

Gastric ulcers, if small and showing a tendency to heal, can with a reasonable degree of safety be treated medically, in an intensive manner, for a reasonable length of time. If they do not heal, resection or excision should be performed. The large callous ulcer, it is felt, may be on a cancer base and extensive resection is probably always advisable.

There is no doubt that for obstructing duodenal ulcers in the presence of low acidity, gastro-enterostomy is the safest and best surgical procedure that can be performed. Gastro-enterostomy can likewise be used to great advantage in the treatment of many large ulcers of the perforating type. Gastric acidity can be satisfactorily controlled in the majority of cases by the dilution and neutralization of the acid gastric secretion brought about by regurgitated jejunal content. Relative achlorhydria is brought about in a goodly percentage of cases in which gastro-enterostomy has been performed.

Nevertheless, surgeons have been looking for a more satisfactory method of treating duodenal ulcers than by gastro-enterostomy. At the present time, the method of gastric resection is receiving great attention. That in selected cases this operation gives excellent results is beyond question. Lahey[8] has said that there seems little question but that the best surgical results both immediate and remote are in those ulcer patients who postoperatively have a low acidity or anacidity, and that the operation which most consistently brings this about is extensive gastrectomy.

When entero-anastomosis is performed—unless the opening is quite small—after gastro-enterostomy, it naturally tends to divert the duodenal secretion into the jejunum rather than directly into the stoma of the gastro-enterostomy and, therefore, predisposes to the development of jejunal ulcer.

The ideal result after gastro-enterostomy or gastric resection is in those cases in which the stoma between the stomach and the bowel tends to function naturally and the food remains in the stomach long enough for digestion to get well started. The so-called "dumping" stomach, which empties its contents into the jejunum a short time after ingestion may interfere seriously with normal digestion at times and may cause, in some cases, a feeling of faintness, abdominal distress, *etc.*

In cases in which there is a stomach ulcer high up on the lesser curvature, a greater portion of the stomach can be conserved by making the resection in the form of a modified right angle or L, a groove, slot, or V wedge being taken

out of the lesser curvature up to and including the ulcer and allowing the transverse section of the stomach to be made a little farther down toward the antrum than would be necessary otherwise. In gastric resection, the Hofmeister and similar resection technics tend to preserve the functions of the stomach as such and to slow down, to some extent, the emptying time of the remainder of the organ. In cases of large callous ulcer, when favorably situated, we have usually resorted to stomach resection.

The general surgical trend during the past several years has been toward medical treatment for most duodenal ulcer cases.

A tabulation of the findings of the roentgenologist has been included, to show the comparative incidence of peptic ulcers in this section of the country (Table I).

TABLE I

PATHOLOGIC FINDINGS IN STOMACHS EXAMINED ROENTGENOLOGICALLY

April, 1934, to November, 1938

Lesions of the Esophagus:	
Cardiospasm or achalasia of the cardia	2
Carcinoma	3
Congenital atresia	2
Benign stricture	2
Syphilis	1
Varices	1
Diverticuli	1
Lesions of the Stomach:	
Ulcer (benign)	12
Carcinoma	38
Suggestive gastritis	20
Polypi (one or more)	2
Syphilis	1
Lesions of the Duodenum:	
Ulcers	281
Diverticuli	12
Obstruction from extrinsic lesion	6
Carcinoma, primary (1); secondary pancreas (1)	2
Hypertrophy of the pyloric muscle	4
Gastrojejunal ulcers	3

A review of cases having stomach symptoms that have been examined at the clinic of the Charleston General Hospital by fluoroscopy during the past few years has been shown in Table II.

TABLE II

SYNOPSIS OF FINDINGS IN STOMACHS EXAMINED FLUOROSCOPICALLY

From April 1, 1934, to December 1, 1938

Total stomachs examined	1,688
Negative stomach findings	1,236
Pathology found	409
Miscellaneous—reexamined, unsatisfactory, patient not properly prepared, *etc.*	92

In the month of December, 1938, there were 40 stomachs examined roentgenologically, which showed negative findings in 24, duodenal ulcers in 13, and miscellaneous, three.

ILLUSTRATIVE CASE REPORTS

Case 1.—J. K., white, male, age 54, was admitted to the Charleston General Hospital, May 23, 1936, complaining of pain in the right side. In 1932, patient first noticed a lump under the right costal margin. This was slightly tender but not painful and he paid no attention to it. No symptoms attributable to disturbances of the gastro-intestinal tract had been noted by the patient. Two weeks previously, he had developed an aching pain with some swelling in the right lower quadrant, associated with anorexia and occasional moderate vomiting episodes. Rest in bed for four days eased the aching somewhat but it did not disappear. The symptoms continued without much change except diminution of vomiting during the past week.

Physical Examination showed a cachectic man with a pale, dry, parched skin. The right epitrochlear node was palpable. Blood pressure 122/70. Temperature 100.6° F. The chest was filled with asthmatic squeaks. The liver was markedly enlarged and extended two fingers below the umbilicus; its surface and edge were smooth. There were both tenderness and spasm in the right lower quadrant, right upper quadrant, right flank and right costovertebral area. Hemoglobin 70 per cent, leukocytes 18,600; Kline negative. Uranalysis negative.

Roentgenologic examination showed a considerable amount of opaque material over the left side of the abdomen and in the left pelvis. There was also apparent a soft tissue tumor in the right upper quadrant which was seemingly separate from the right kidney. On May 30, 1936, seven days later, another roentgenologic examination was made which showed a considerable deformity of the duodenum with a perforating ulcer on the greater curvature and posterior wall.

Operation.—June 2, 1936: Through an upper right rectus incision, a duodenal ulcer was found which had perforated the posterior wall of the duodenum over the pancreas. The perforation had burrowed in horseshoe-fashion upward alongside the gallbladder through the edge of the liver and about halfway through the abdominal wall. After dissecting the sinus down to the level of the duodenum, it was found that the surrounding tissues were so friable that it was thought safer to plug the opening with omentum rather than to undertake its closure by suture. In addition to this a posterior gastro-enterostomy, with a wide stoma, was performed.

Subsequent Course.—The patient made an uneventful recovery following the operation, and at the time of discharge, June 30, 1936, his condition was greatly improved. Subsequent roentgenologic examination after six months, and again two years later, found the duodenum to be in fair condition, with no evidence of a crater.

Case 2.—F. S., white, female, age 52, was admitted to the Charleston General Hospital, complaining of generalized abdominal pain, nausea and vomiting, loss of weight and dysuria. The patient had been troubled for the past five years with indigestion. She often had nausea and vomiting, and pain in the epigastric region. These symptoms had gradually become more severe. She had lost considerable weight. She said that about five years ago she had had a roentgenologic examination of the stomach, and was told that she had an ulcer. Roentgenologic examination, March 30, 1935, showed a perforated ulcer at the junction of the middle and distal thirds of the stomach on the lesser curvature. The base was nearly 2 cm. in diameter with moderate induration in this area. The duodenum was negative. Hemoglobin 87 per cent; leukocytes 9,300; neutrophils 64 per cent; erythrocytes 4,360,000. Uranalysis showed 2 plus pus, specific gravity 1.002.

Operation.—April 4, 1935: The antral portion of the stomach was resected, and a

gastrojejunostomy of the Pólya type was performed. An ulcer was found on the lesser curvature of the stomach, which measured about 1.5 cm. in diameter.

Subsequent Course.—The patient had a normal convalescence and a short time later was checked roentgenologically, which showed satisfactory results. On January 7, 1937, a second examination showed her stomach to be functioning normally and no evidence of disease.

Case 3.—D. S., white, female, age 40, was admitted to the Charleston General Hospital, November 30, 1936, complaining of having had stomach trouble for many years. It showed improvement at times and then got worse. The symptoms have become more severe and she has had an attack of two months' duration. Has had considerable pain at times. Has not had much nausea or vomiting. The chief complaint was pain in the epigastrium reflected to the left side. Patient had vomited blood in the past but not recently.

Roentgenologic Examination.—Dr. V. L. Peterson: "There is a perforating ulcer on the posterior wall, near the lesser curvature of the stomach, at the junction of the proximal and middle thirds. There is a palpable tumor within the stomach as well as a palpable mass, extragastric. This would indicate a malignant condition rather than a benign ulcer. However, the mass could be inflammatory tissue about a chronic ulcer. I believe it should be explored surgically and examined pathologically."

Operation.—December 2, 1936: Approximately one-half of the stomach containing a mass, about two inches in diameter, on the posterior surface was resected. There were a number of adhesions on the posterior surface of the stomach probably due to chronic perforation of an old healed ulcer. After resection, an end-to-side anastomosis was made with the jejunum, posterior Pólya type. *Postoperative Diagnosis:* "Polyposis of stomach, occurring around the edges of an old healed ulcer. There are four polypi, each approximately two-thirds of an inch in diameter."

Subsequent Course.—Patient made apparently satisfactory progress for several days, then developed bronchopneumonia and died, December 11, 1936.

Autopsy.—It was found that the suture lines of the stomach and small bowel were intact and that there was no evidence of peritonitis. The final cause of death was stated to be: "Purulent bronchitis and bronchiolitis with early bronchopneumonia."

Case 4.—D. C., white, female, married, age 68, was admitted to the Charleston General Hospital, May 12, 1838.

Roentgenologic Examination.—Doctor Peterson: "There is a marked gastric retention with a penetrating ulcer on the lesser curvature of the stomach at about the juncture of the middle and distal thirds. The entire ulcerated area measures approximately 7 cm. in diameter. This should be considered a malignant gastric ulcer. Unable to visualize the duodenum because of the obstruction. This may be secondary to a perigastritis about the penetrating ulcer. There is a normally functioning gallbladder."

Laboratory Data.—Hemoglobin 75 per cent; erythrocytes 4,500,000; leukocytes 11,000; lymphocytes 26 per cent; monocytes 4 per cent; neutrophils 70 per cent. Uranalysis negative except for a very occasional pus cell. Weight 70 pounds.

Preoperative Diagnosis.—Malignant, perforated ulcer of the lesser curvature of the stomach.

Operation.—May 14, 1938: A large ulcer mass was noticed on the lesser curvature of the stomach which measured approximately two and one-half inches across. It was deeply perforated in the center. The gallbladder and pelvic organs were normal. Gastric resection of approximately one-half of the stomach, and an end-to-side gastrojejunostomy were performed.

Pathologic Examination.—Doctor Putschar: "Large ulcus callosum of the lesser curvature with very marked hypertrophic gastritis."

Subsequent Course.—Patient made a satisfactory recovery from this operation and was discharged, May 22, 1938. She has been getting along in a satisfactory manner and

has gained weight until now she weighs 140 pounds. A recent roentgenologic study indicated that the stomach was functioning satisfactorily.

Case 5.—H. D., white, male, age 62, was admitted to the Charleston General Hospital, April 5, 1937, complaining of pain in the epigastrium, gas in stomach and constipation. The onset of his symptoms was about two years before, when he began to be troubled with constipation, had a good deal of gas in stomach, belched considerably and had some pain in the epigastrium. This pain usually was relieved by food or taking of alkalies. His symptoms had no relation to greasy foods. He had never had any severe attack of pain or jaundice. He had not vomited or noted any tarry stools. He had been told that he had chronic gallbladder trouble with stones.

Physical Examination was not remarkable except that he was somewhat tender in the epigastrium. Roentgenologic examination by Doctor Peterson showed a rather marked deformity of the duodenum apparently due to an active ulcer. *Laboratory Data:* Hemoglobin was 84 per cent; erythrocytes 4,700,000; leukocytes 7,200, 74 per cent neutrophils, 23 per cent lymphocytes, 2 per cent monocytes, 1 per cent eosinophils. Uranalysis negative. Kline test negative. *Clinical Diagnosis:* Duodenal ulcer.

Operation.—April 7, 1937: A posterior gastro-enterostomy was performed. He was also found to have a solitary gallstone in the gallbladder. Cholecystectomy and appendicectomy were also performed.

Subsequent Course.—Postoperative recovery was satisfactory except that he had a moderate wound infection. This cleared up rather promptly; however, he later developed an incisional hernia.

He was relieved of his symptoms for a time but they recurred and he returned to the Hospital, September 11, 1938, complaining of pain in the epigastrium, belching of gas, *etc.* These symptoms were relieved by alkalies and food. Roentgenologic examination again showed the presence of a duodenal ulcer.

It was thought that his symptoms would probably be relieved if a division of the stomach were performed, and on September 13, 1938, the pyloric end of the stomach was divided, both ends were closed, and repair of the incisional hernia was effected with chromic catgut and fascia lata sutures. He recovered satisfactorily and was discharged, October 1, 1938. The results have been fairly satisfactory.

Case 6.—L. N., white, male, age 36, was admitted to the Charleston General Hospital, January 3, 1939, complaining of vomiting and gastric pain. About one year ago, he had his first attack and has had several mild episodes since. It was his opinion that greasy foods brought on these attacks of vomiting because when he ate foods that were not greasy, he was not troubled so much. The present attack started about one month ago, and since that time he has continued to vomit practically everything taken by mouth. He became very emaciated, weak and lost much weight during the past month. He had not vomited any blood at any time. Patient did not give any previous history of pain in the stomach, tarry stools or other usual symptoms of peptic ulcer.

Physical Examination was essentially negative, other than for his evident emaciation. *Laboratory Data:* Hemoglobin 78.4 per cent; erythrocytes 4,500,000; leukocytes 12,000, 24 per cent lymphocytes, 76 per cent neutrophils. Uranalysis negative. Wassermann 4 plus. Gastric analysis was essentially normal as to the amount of acid.

Roentgenologic examination of his stomach revealed practically complete obstruction at the pyloric end. A definite lesion as to ulcer could not be made out and the exact nature of this obstruction could not be diagnosed from the roentgenograms, except that there was an obstructive lesion at the pyloric end of the stomach.

Operation.—January 9, 1939: A great deal of edema in the region of the pyloric end of the stomach and mostly in the posterior wall was found. On opening the stomach, no definite ulcer could be identified. The pyloric opening was almost closed, and two slender adhesion bands were noted crossing the opening. There was definite thickening and edema of the antral end of the stomach wall, which involved the posterior

wall for the most part. Subtotal resection of the stomach and an end-to-side gastrojejunostomy were performed.

Pathologic Examination.—Inflammatory stenosis of the pylorus with small ulceration and adhesions. There was an associated diffuse, chronic gastritis and edema.

Subsequent Course.—His postoperative course was very satisfactory. He was discharged on the fifteenth postoperative day.

Case 7.—G. M., white, male, age 63, was admitted to the Charleston General Hospital, June 24, 1937, complaining of severe pain and tenderness in the upper part of the abdomen. For the past seven years, he had been having symptoms of peptic ulcer. He stated that about three days before admission as he was arising from bed, he had a sudden, severe attack of pain in the region of the epigastrium. This pain was rather generalized over the abdomen and after a short intermission, he continued to have the same type of pain. At the time of admission he experienced pain as he moved his body in any way or had pressure made on his abdomen.

Physical Examination revealed an elderly, emaciated man, with a large head, who looked acutely ill. He was exquisitely tender and rigid over the upper part of the abdomen and complained of severe pain on slight motion of his body. A flat roentgenogram of the abdomen, in the upright position, did not show any abnormality of the diaphragm but did show extensive fibrocystic changes in the spine, pelvis and both femora, which, apparently, were due to Paget's disease. *Laboratory Data:* Hemoglobin 85 per cent; erythrocytes 5,000,000; leukocytes 12,400; 81 per cent neutrophils, 16 per cent lymphocytes, 3 per cent monocytes. Kline test was negative.

The patient was thought to have either acute cholecystitis or a perforated duodenal ulcer which had become walled-off. For this reason, he was treated symptomatically for a few days. On the fourth day after admission he began to vomit and show signs of obstruction at the pyloric end of the stomach. It was thought advisable to perform an exploratory operation.

Operation.—June 29, 1937: Local anesthesia. There was a marked inflammatory condition about the pyloric end of the stomach and it was thought that he had a perforated ulcer which was walled-off. Posterior gastro-enterostomy was performed.

Subsequent Course.—He made a very satisfactory recovery from the operation. His stomach was checked fluoroscopically, after he had recovered sufficiently, which revealed an obstruction at the pyloric end of the stomach which Doctor Peterson thought might possibly be malignant but at the time of operation, it looked more like a perforated ulcer. The gastro-enterostomy was working satisfactorily. His family physician has recently reported that this patient has continued to do well.

Case 8.—E. B., white, male, was admitted to the Charleston General Hospital, June 14, 1935, complaining of epigastric pain. There had been some aching over the epigastrium for two weeks. He had not vomited or been nauseated at any time. Food seemed to relieve his discomfort. Had had indigestion, sour stomach, pyrosis and gas even since he could remember. During childhood he had an attack of pain similar to the one that had been present for the last two weeks. Once he was put on a diet for ulcer of the stomach and as it did not seem to give him much relief, he discontinued it after two months. Had had chronic constipation; also had had occasional mild burning on urination for the past five years.

Laboratory Data: Hemoglobin 84 per cent; leukocytes 12,500, 85 per cent neutrophils; Kline test negative; uranalysis negative.

Operation.—June 14, 1935: A small ulcer was found on the anterior surface of the first portion of the duodenum, surrounded by localized peritonitis with a few flecks of fibrinous exudate on the bowel immediately surrounding the perforation. The perforation was closed in the usual manner and reinforced with an omental patch.

Postoperative course was uneventful and patient was allowed to go home on the twelfth day postoperative, at which time he was feeling quite well.

Case 9.—J. C., white, male, age 40, was admitted to the Charleston General Hospital, April 5, 1936, complaining of pain in the midepigastrium.

He had had indigestion for the past two or three days, and the day before admission to the hospital, was suddenly seized with generalized, acute abdominal pain that was not relieved by a hypodermic of morphine. The day of admission, the generalized pain in the abdomen became very severe.

Physical Examination revealed an acutely ill patient. Temperature 102.6° F.; pulse 120. There was generalized, board-like rigidity of the abdomen with the point of maximum tenderness just to the right of the umbilicus. *Laboratory Data:* Leukocytes 15,000, 82 per cent neutrophils. Uranalysis negative except for a trace of albumin. Kline test negative.

Operation.—April 5, 1936: The peritoneal cavity was found filled with a milky fluid and there was a diffuse peritonitis. A small, perforated ulcer was found on the anterior surface of the duodenum, which was sutured with black silk. The abdominal cavity was irrigated with saline solution. Patient did fairly well for 24 hours after operation but continued to run a high temperature. The course was downhill, however, and he died, April 8, 1936. No autopsy was obtained.

Case 10.—D. S., white, female, age 49, was admitted to the Charleston General Hospital, August 6, 1936, complaining of pain in the epigastrium and right upper quadrant, swelling of feet and ankles, shortness of breath and backache. Twelve years ago, she had been operated upon, elsewhere, at which time a gastro-enterostomy had been performed for the relief of a duodenal ulcer. She said that she had been comparatively comfortable up until a few months before coming to the Hospital.

The patient stated that several days prior to the date of admission, she suddenly developed a sharp, shooting pain which started under the right costal margin and radiated through to the right scapular region. After a short while the pain became constant in character. She had had clay-colored stools for about one week and was jaundiced. She thinks that she had passed some tarry stools lately. She had been told that she had high blood pressure. After being on her feet for any length of time, she noticed that they became moderately swollen. She had moderate dyspnea on exertion and had been bothered by palpitation of the heart.

Physical Examination revealed a somewhat emaciated patient, who appeared to be moderately ill. Temperature, pulse and respiration were normal. Blood pressure 120/74. The examination was otherwise negative except for tenderness in the right upper quadrant of the abdomen and in the midlumbar region. There were no palpable masses in the abdomen. There was moderate rigidity of the muscles over the right upper quadrant and in the epigastrium. *Laboratory Data:* Leukocytes 10,950, 82 per cent neutrophils. Uranalysis negative. *Clinical Diagnosis:* Recurrent peptic ulcer and possibly cholecystitis. Roentgenologic examination showed the presence of a good-sized duodenal ulcer.

Operation.—The gastro-enterostomy was taken down, and the gastrojejunal ulcer excised. The continuity of both jejunum and stomach was restored, and a Judd pyloroplasty was performed.

Subsequent Course.—The patient made a good operative recovery, but within a few weeks she began to complain of symptoms incidental to peptic ulcer. After this, she was treated medically for some time, but without avail; finally it was decided that the only thing to do would be to resect the ulcer in the duodenum and a considerable portion of the stomach.

Second Operation.—It was found that there was not only evidence of chronic gastritis but that there was a great deal of scar tissue formation with superficial ulceration of the duodenum adjacent to the pylorus. The first portion of the duodenum and a little more than one-half of the stomach were resected.

Subsequent Course.—The postoperative course was uncomplicated except for an attack of diarrhea which was soon controlled. She was discharged from the Hospital,

September 3, 1936, entirely free from discomfort. A follow-up, several months later, indicated that she had complete relief.

Case 11.—C. P., white, female, married, age 33, was admitted to the Hospital, February 14, 1938, complaining of pain in the epigastrium and right side, and vomiting, since November, 1937. She had had a cholecystectomy performed two years before. For three to four months postoperative, she had severe nausea and vomiting. Was well until November, 1937, when she again developed nausea and vomiting, and pain in epigastrium and right hypochondrium, which have persisted ever since. "Seems like a rock in her stomach."

Physical Examination revealed a patient, underweight and undernourished. There was tenderness in the epigastrium. *Laboratory Data:* Uranalysis essentially negative except for an occasional renal cell and 1 plus pus. Hemoglobin 65 per cent; erythrocytes 4,400,000; leukocytes 9,200, 24 per cent lymphocytes, 2 per cent monocytes, 74 per cent neutrophils; Kline test negative. Fractional test meals showed: One hour, showed a total acidity of 53 degrees with 34 degrees free HCl. One and one-quarter hours, 14 degrees total acidity, six degrees free HCl. One and one-half hours, 9 degrees total acidity and no free HCl. One and three-quarters hours, ten degrees total acidity and no free HCl. Two hours, 13 degrees total acidity and no free HCl.

Roentgenologic examination showed there to be a marked deformity of the duodenum with a crater present—indicating an active duodenal ulcer. There was also a definite retention of barium in the stomach. *Clinical Diagnosis:* Peptic ulcer with gastric retention.

Operation.—February 17, 1938: The antral end of the stomach and pylorus were found to be densely adherent, in part, to the undersurface of the liver and, in part, to the anterior parietal peritoneum. This condition had apparently resulted from the removal of the gallbladder two years before. The pelvis and uterus were essentially normal except for the presence of a few adhesions. A posterior gastro-enterostomy, using the usual no-clamp technic, was performed.

Subsequent Course.—Roentgenologic examination, February 26, 1938, showed, after a small amount of barium had been instilled into the stomach through a Levine tube, that it was discharged through the duodenum and passed by the stoma of the gastro-enterostomy into the distal loop of the jejunum, which phenomenon is not unusual following a recent operation. The patient's condition on discharge was satisfactory. Recent advices from her physician state that she has been well and comfortable up to the present time.

Case 12.—A. G., white, female, married, age 58, was admitted to the Charleston General Hospital, May 22, 1938, complaining of a good deal of gastric pain and discomfort, which had been present for some years. In 1917, she had had a posterior gastro-enterostomy performed, which had helped her for a time, but later on the pain returned. She had had at another time a thyroidectomy; also, on another occasion, a pelvic operation, the details of which were not known.

Roentgenologic examination, May 18, 1938, showed a functioning gastro-enterostomy, and no evidence of ulceration in this area. There was a deformity of the duodenum with a marked tenderness—indicating a recurrence of the old duodenal ulcer. *Laboratory Data:* Uranalysis essentially negative except for an occasional pus cell. Hemoglobin 71.5 per cent; erythrocytes 4,700,000; leukocytes 4,600, 68 per cent lymphocytes, 2 per cent monocytes, 28 per cent neutrophils, 2 per cent eosinophils; Kline test negative. Inasmuch as it was felt that this patient had a duodenal ulcer, it was felt that the condition could be cured by an exclusion procedure.

Operation.—The antral end of the stomach was divided a short distance above the pyloric sphincter and both ends closed. The gastro-enterostomy was examined at the time of operation and found to be in satisfactory condition.

Subsequent Course.—The patient made a satisfactory convalescence, but, after dis-

charge, she had a good deal of stomach disturbance for about three months. More recently, her physician reports that she has been well and comfortable for some time.

Case 13.—E. T., white, female, married, age 69, was admitted to the Charleston General Hospital, March 17, 1935, complaining of tenderness over the epigastrium. Since December, 1935, the patient had been suffering from pains in the abdomen and under the ribs. She vomited frequently. Took laxatives and her stools had been tarry. Lost much weight and her appetite had been quite poor. She had been in the Hospital the year before, at which time she was treated for asthma. Four days before admission, she had a severe hemorrhage from the stomach, the blood was bright red and was accompanied by pain.

Physical Examination.—The patient was in poor general health and nutrition. There was poor first sound of the heart followed by an early diastolic or late systolic, rough murmur. The abdomen had very poor muscle tone and was scaphoid. A mass could be palpated in the upper abdomen just to the right of the midline. There was some tenderness over the upper right quadrant and epigastrium. The liver dulness hardly reached the fifth rib, and its edge was not felt. Hemoglobin 81 per cent; erythrocytes 7,600,000; Kline test negative.

Roentgenologic Examination.—"Ulcerating carcinoma involving the distal third of the stomach. I would like to re-ray to check the cardia." The second report was: "There are multiple areas of ulceration throughout the midportion of the stomach, with narrowing and loss of rugae folds. The lesion involves the entire stomach." Roentgenologically it was considered malignant, although a mass could not be palpated through the abdominal wall. Syphilis was suggested, and it was asked that this be ruled out before operation.

Operation.—March 23, 1935: On opening the stomach, it was found that numerous ulcerated areas of mucosa could be seen. It was also noted that the stomach wall was about three times its normal thickness. There were numerous ulcerated areas on the greater curvature as well as elsewhere. In view of the history and pathology noted, the diagnosis was made of chronic gastritis with multiple ulcers of the stomach. A subtotal resection of the stomach was performed, leaving approximately two-thirds of the stomach, and an anastomosis between the jejunum and resected end of the stomach effected. *Pathologic Report:* "Acute ulcerative gastritis superimposed upon a chronic gastritis."

Subsequent Course.—The patient made a satisfactory recovery and was seen and checked several times during the next two years. She had gained in weight, looked well, and said that she had been entirely relieved of her previous symptoms.

Case 14.—R. K., white, male, age 27, was admitted to the Charleston General Hospital, July 9, 1935, complaining of pain in the stomach. The patient stated that for the past five or six years he had had an acid stomach and had taken antacid powders, *etc.*; at times he had sharp pains in the epigastric region which were relieved by taking food. The condition grew progressively worse, and about nine months ago, he had a hemorrhage, following which he was taken to the Hospital, where a diagnosis of duodenal ulcer was made. He remained for 15 days, and subsequently got along fairly well until about two weeks ago when he again started to have pain. This had grown much worse, and for the past three days, it was not relieved by food or medication. He had not vomited but the stomach seemed to be filled with gas. No loss of weight was noted.

Physical Examination.—The patient was well developed and well nourished. There was a mild rigidity and spasm over the epigastric region with tenderness to deep palpation over the upper abdomen. Gastric analysis showed free HCl of 45 degrees, total acidity 70 degrees; erythrocytes 5,250,000; hemoglobin 90 per cent. Kline and Wassermann tests were negative. The roentgenographic findings were interpreted as showing an "ulcerating carcinoma on the greater curvature involving the pyloric end of the stomach."

Operation.—July 16, 1935: A large ulcer was found on the greater curvature of the

stomach which caused a great deal of deformity, and which was covered with adherent omentum. A Billroth-Pólya type of operation was performed, resecting the pylorus and lower half of the stomach.

Pathologic Report.—Acute and chronic gastric ulcer (greater curvature). Carcinoma, probably Grade 4, arising from the glandular epithelium.

Subsequent Course.—In spite of the youth of this patient, his prognosis was considered hopeful for a permanent cure. The patient made a fairly uneventful recovery and left the Hospital on the fourteenth postoperative day, feeling stronger and able to take nourishment well, and was entirely free from gastric discomfort. A report from his physician, December 5, 1936, stated that he was in good physical condition and the results of treatment had been very satisfactory. Later, roentgenologic examination showed a normally functioning stoma with no signs of recurrence. A recent check-up by his physician indicates the continuance of good health.

COMMENT.—In Case 1 (J. K.), the tissues surrounding the perforation had been so badly damaged by inflammatory reaction that it was thought best to close this large perforation by plugging it with omentum, rather than to attempt to close it by suture. The immediate results were good and subsequent roentgenologic examination, made some months later, showed that healing had taken place.[5] Dr. Waltman Walters[6] recently reported a somewhat similar case, successfully handled by the same method.

Case 10 (D. S.) illustrates the fact that no one surgical procedure may bring about a permanent cure. This patient stated that, 12 years previously, she had been operated upon, elsewhere, for the relief of duodenal ulcer. At this time, a gastro-enterostomy was performed. Following this she was quite comfortable for nearly 12 years. She then began to suffer from sharp pain which usually started under the right costal margin and radiated through to the right scapular region. Later on this pain became constant in character. Roentgenologic examination revealed a jejunal ulcer. The ulcer was excised, the gastro-enterostomy taken down, and a Judd pyloroplasty was performed. Within a few months she was back at the Hospital. Roentgenologic examination showed a duodenal ulcer. At this time, it was thought advisable to treat the condition radically. The ulcer was excised and about one-half of the stomach was resected, followed by an end-to-side anastomosis with the jejunum. This patient's condition was relieved, and so far has remained good.

Case 14 (R. K.) emphasizes the long history of stomach disorder that so often indicates an ulcer; also the fact that it is often impossible to distinguish between ulcer of the stomach and an ulcerating carcinoma, except roentgenologically, especially in the earlier stages.

CONCLUSIONS

The problems of digestive acid control and tissue susceptibility are still unsolved and can be met only in part. The great majority of duodenal ulcers can be treated medically with satisfactory results, but a comparatively small percentage of duodenal ulcers that are progressive in nature, that are prone to bleed, and that are resistant to treatment may require surgery. When, owing to adverse economic conditions, these patients are unable to carry out

adequate medical and dietetic regimens, surgical intervention may become necessary.

Owing to the danger of malignancy, all gastric ulcers that are large or have a definite crater, also the smaller gastric ulcers that fail to heal under adequate medical treatment, call for surgical treatment by excision or resection.

Gastro-enterostomy is an eminently satisfactory treatment for the chronic duodenal ulcer associated with low gastric acidity.

Excessive gastric acidity and hypersecretion can best be controlled by gastric resection.

The pylorus and antral end of the stomach probably are, to some extent, responsible for the secretion of gastric acid.

Postoperative care of ulcer cases is most important, and, for the successful treatment of peptic ulcers, requires the close cooperation of internist, roentgenologist and surgeon.

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PRIMARY CARCINOMA OF THE ILEUM

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In 41,848 autopsies performed in the Vienna General Hospital, Johnson found 343 cases of carcinoma of the intestine, of which ten were in the ileum. In 21,624 autopsies, Brill found 50 cases of carcinoma of the ileum. In 20,480 autopsies, reported by Maydl, there were 100 cases of carcinoma of the intestine, of which four were in the ileum. These combined statistics would indicate that in a large series of routine postmortem examinations, carcinoma of the intestine was found in 0.7 per cent and carcinoma of the ileum was found in 0.0075 per cent (75/10,000 of 1 per cent). Viewed from another angle, Ewing states that of the intestinal cancers 8.58 per cent arise in the small bowel and more than half of these in the duodenum. The Mayo Clinic, with its vast number of abdominal operations, reports 69 cases of carcinoma of the small bowel, of which 14 were in the ileum. Of these 69, 47 were in males and 22 in females. Carcinoma of the ileum, while not rare, is very infrequent.

Case Report.—M. H., white, female, single, age 37, was admitted to the hospital, October 17, 1937, complaining that during the previous two or three months the slightest exertion tired her, and it was with increasing difficulty that she could perform her duties as a teacher. During the two to three weeks prior to admission she had to "drag herself around." She thought she had lost some weight but was sure it was not very much. She came to the hospital to determine the cause of her increasing weakness. The only other symptom was vague abdominal discomfort with considerable gas. Her family and past histories were essentially negative.

Physical Examination.—The patient was definitely anemic. Heart, lungs and extremities normal. On deep palpation over the lower abdomen there was slight tenderness and a vague suggestion of an indefinite, elusive soft mass. Vaginal and rectal examinations were negative. Temperature, 100.4° F.; pulse, 120; respiration, 28.

Laboratory Data.—Urine normal. Blood examination: R.B.C. 2,840,000, Hb. 32 per cent; W.B.C. 11,450, polys 71 per cent, small mononuclears 24 per cent, large mononuclears 3 per cent, transitionals 1 per cent, eosinophils 1 per cent. Sedimentation rate, at 15-minute intervals, 31, 34, 37 and 38 Mm., respectively. Four days later the red count had dropped from 2,840,000 to 2,020,000 and the hemoglobin from 32 to 30 per cent. Blood cultures were negative.

Test meal showed total acidity 65, free HCl 30, many starch granules and yeast cells, no bile, occult blood or lactic acid. Much mucus. Examination of feces for occult blood strongly positive.

Roentgenologic Examination.—Dr. G. W. Murphy: A barium enema showed the entire colon well filled and quite irritable. Some redundancy of descending and transverse colon. No filling defect or changes in wall seen. Lungs negative. Left ventricle of heart moderately enlarged. No organic lesion seen in stomach and duodenum. A barium meal showed the stomach empty at the sixth hour. Some 24-hour retention of enema. At 24 hours, no sign of obstruction. Appendix freely movable, not tender and well filled. Only significant finding is the cardiac enlargement. (Later on, with the full knowledge of the nature and location of the disease, a review of the roentgenograms still gave no clue to the diagnosis, other than to exonerate the stomach and the large bowel.)

Confirmatory Physical Examination.—Dr. Walter Johnson: "Pallid complexion, otherwise healthy appearing woman, age 37. Well developed and well nourished. Eyes normal except anemic. Nose and throat normal except pallid mucosa. Nodes not felt.

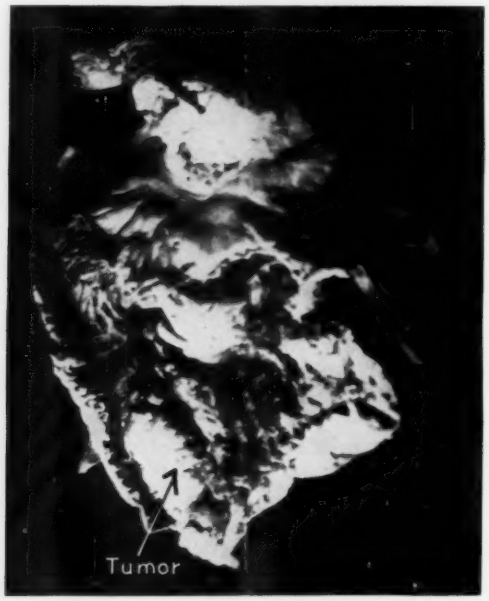

FIG. 1.—Photograph of the gross specimen. The tumor is indicated by the arrow.

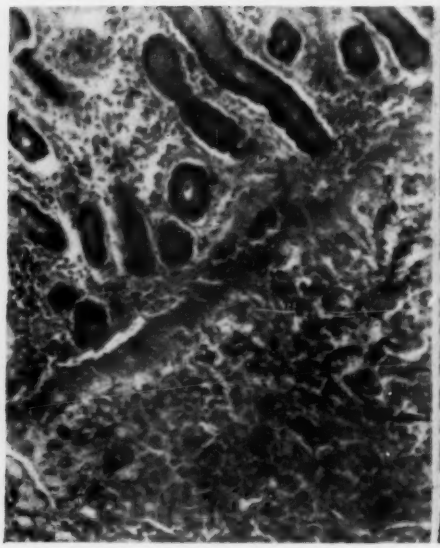

FIG. 2.—Photomicrograph showing the glandular mucosa, and underneath the muscularis mucosa is the tumor tissue—an adenocarcinoma of the ileum.

FIG. 3.—Photomicrograph showing the densely packed glandular deposits which constitute the neoplasm.

Chest normal. Pulse 120, pulmonic sounds roughened. Blood pressure 120/84. Abdomen moderately tense and fairly obese. Indefinite midline mass above navel? Reflexes normal."

Discussion.—The marked asthenia, the severe secondary anemia, the slight loss of weight, together with the suspicion of an elusive mass in the abdomen, suggested a probable diagnosis of malignancy somewhere in the abdomen. The occult blood in the feces indicated the gastro-intestinal tract. The stomach, the first part of the duodenum, and the large bowel had been excluded with reasonable certainty. Thus by a process of elimination, a probable diagnosis of carcinoma of the small intestine was arrived at.

Obviously, the patient was in no condition for operation, but donors were obtained for blood transfusion in the hope that a celiotomy might be performed later. At this stage, October 22, 1937, the patient decided to return to her home in Morristown, Tenn., and was referred to Dr. L. W. Nabers of that city. She died, November 15, 1937, less than one month after we first saw her.

Autopsy.—Dr. L. W. Nabers: "There was a large, perforated mass of ileum, about three feet from the ileocecal junction, and its large size and great weight caused it to fall into the pelvis where it perforated into the posterior cul-de-sac. *Grossly,* the tumor was very cellular, in fact, necrotic in places. There were multiple metastases to the liver, none to the lungs. Examination of the heart negative."

Pathologic Examination.—Dr. Murray M. Copeland, Baltimore: "The specimen of bowel tumor shows on section a rather diffuse small cell type of carcinoma arising from the mucosa of the small intestine, invading the musculature of the bowel. The lymph nodes show metastases of the same type of carcinoma. My impression is that this is an adenocarcinoma of the small bowel, Grade 4, with metastases."

An independent pathologic report by Dr. Alfred Blumberg, Asheville, was: "*Gross:* The ulcerated area is 14 cm. long. When opened it contained a shaggy mass, white to gray in color. The white color was due to tumors varying in size, the largest being 5 cm. long, 2 cm. wide, and 2 cm. thick. In addition, mesenteric lymph nodes had undergone cancerous changes. These lymph nodes are white in color and somewhat mushy in the center, otherwise they are of the same appearance as are the tumors within the ileum. *Microscopic.*—In certain areas a quite distinct glandular mucosa is seen in which carcinomatous nodules are present. The neoplasm interrupts the muscularis mucosa and continues its course beneath it. The agminated follicles are replaced by the neoplasm which is quite extensive. In some areas ulceration had taken place. The neoplasm is adenocarcinomatous in character, quite vascular, and supported by a stroma which is often very delicate. Infiltration of neoplastic tissue into veins is not unusual and necrotic changes are present in numerous places. *Pathologic Diagnosis:* Adenocarcinoma of ileum.'"

There are no pathognomonic signs or symptoms of early carcinoma of the small bowel. Loss of weight, asthenia, anemia, indigestion, anorexia, flatulence and vague abdominal symptoms may occur in various combinations and degrees. Melena is rare, but occult blood in the feces is frequently present. Owing to the semifluid contents of the small bowel, by the time the disease has produced symptoms of obstruction, it is nearing the terminal stage.

The prognosis is poor. If discovered early, resection is indicated. After symptoms of obstruction have developed, anastomosis around the growth with a lower primary mortality would probably be better judgment than to attempt resection with a high immediate mortality and with such slight chance of a cure.

In the Rankin and Mayo series of carcinomata of the small intestine, no patient lived longer than three years after the diagnosis, with an average duration of life of less than one year. The object of the present communication is: (1) To put on record one more authentic case of carcinoma of the ileum. (2) To emphasize the fact that carcinoma of the small bowel is insidious in its

development, and that only in the late and practically hopeless stage does it give signs or symptoms sufficient to arouse even a suspicion of its nature. (3) Unless there is obstruction it is almost impossible to make a positive diagnosis of carcinoma of the small intestine. A probable diagnosis may be made by eliminating other lesions, malignant and benign, of the gastro-intestinal tract.

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DISCUSSION.—DR. J. SHELTON HORSLEY (Richmond, Va.): Carcinoma of the small intestine, while rare, is more frequent than it was formerly supposed to be. Occasionally, it is difficult to establish the type of tumor found in the small intestine. Thus, in a patient of mine (Mrs. R. J. B., Path. No. 8938), age 49, there was obstruction with typical symptoms. Operation showed an intussusception about 18 inches from the terminal ileum with a tumor at the apex. The intussusception could not be fully reduced, and the segment of bowel was removed.

On section of the specimen there was a fungus-like growth spread out sharply into the lumen. It was of irregular outline, and friable. Histologic examination showed traces of necrosis with what appeared to be lymphatic tissue or round cell infiltration and rather large clear cells with distinct nuclei. Dr. Joseph McFarland of Philadelphia examined a section and reported as follows: "I frankly don't know what it is, nor do I believe it possible to find out. My impression is that it is of neurogenic origin, and perhaps best regarded as akin to the sympatheticoblastomata. It will be very interesting to see if it ever comes back. I believe that effective eradication frequently results in a cure." Dr. A. C. Broders thinks it is inflammatory granuloma with numerous endothelial leukocytes. Dr. Louisa E. Keasbey, of Lancaster, Pa., believes that the growth is a reticulo-endothelial type of tumor, and quotes Krauspe's article in which he reports several tumors of the same structure. (Krauspe: Verhandl. der Deutsch. Path. Ges., **28**, 166, March, 1935.)

The patient made a satisfactory recovery, and when heard from, October 31, 1938, eight and one-half years after the operation, there was no evidence of recurrence.

Carcinoma of the small intestine is somewhat more frequent in the duodenum, particularly if the length of the duodenum as compared with the rest of the small intestine is taken into consideration. Outside of the duodenum it is more often found in the upper jejunum. An interesting case is that described by Dr. George R. Moffitt, of Harrisburg, Pa. The patient was a female, age 53, who, eight months before admission to the hospital, had had pain in the epigastrium with vomiting, which came on at the same time each day, particularly after eating. The pain was always confined to the epigastrium. There was a feeling of a lump in the abdomen accompanying the attacks of pain. The symptoms increased and became almost constant during the last four weeks before admission to the hospital. Roentgenologic examination showed obstruction in the jejunum. The segment of the jejunum was resected, and an end-to-end union was made, May 11, 1938. The patient made a satisfactory recovery. The neoplasm was a rather malignant type of adenocarcinoma.

Carcinoids are also unusual tumors. They formerly attracted attention as small orange-yellow growths in the tip of the appendix and were thought to be carcinomata. In recent years not a few of these tumors have been reported, located chiefly in the lower small bowel. They spring from a chro-

maffin cell of the intestine, which was first described by Nicholas Kultschitsky, in 1897. In 1905, Schmidt described them again and noted the yellow color. Gosset and Masson, in 1914, added much to the knowledge of these cells, showing that there were granules in the cytoplasm which stained black or brown with silver compounds, and they called them "argentaffin" cells. The origin of these cells is not definitely known, but they are found more frequently in the lower small bowel and in the appendix than elsewhere, in the bases of the intestinal crypts. By some, they are supposed to secrete an endocrine-like substance, as "neurocrine" or adrenalin, but this has not been proved.

These carcinoids or argentaffin tumors are usually benign, but occasionally they become malignant and metastasize into the mesentery and into the liver. Dr. Louisa Keasbey frequently finds them in necropsies, especially in middle-aged or elderly men, as a very small lump in the wall of the small bowel. Apparently they are innocuous and have been present for years.

An unusual feature about these tumors is that histologically the cells of the benign type and of the malignant type are identical, and the diagnosis as to whether the tumor is malignant or benign cannot be established histologically.

A malignant type of this growth is shown by the case of Dr. J. W. Lindsay, of Washington. The patient, white, male, age 56, was admitted to the hospital with a diagnosis of partial intestinal obstruction. A resection was performed and a segment of bowel, about the middle of the small intestine, was removed by Dr. J. O. Warfield, Jr. It had a hard, fibrous contraction. The patient made a satisfactory recovery. Microscopic examination showed the typical appearance of carcinoid. Even when there are metastases in the liver, removal of the original growth seems to prolong life, but most of these are clinically benign.

Primary melanoma of the small intestine has been found in a few instances. Thus, one case by Menne and Beeman (Am. Jour. Dig. Dis. and Nutrit., **3**, 786, 1936–1937) is reported in which there were numerous melanomata of the small intestine; and another case by Lund (New Eng. Jour. Med., **201**, 1133, 1929) seemed to show a primary melanoma. The submucous tissue of the small intestine apparently favors growth of metastatic melanomata.

An interesting secondary melanoma is reported by the late S. W. Budd, of Richmond, and H. C. Jones, of Petersburg. The patient was a white female, age 31, who, in 1930, had a large black mole on her back and a similar one on the arm treated with the electric cautery, but the pigmented scars remained. About two years later, Doctor Jones, on operation, found an intussusception involving the head of the cecum, which had been caused by a pedunculated growth about 18 inches above the ileocecal valve. It was soft and freely movable, and on the peritoneal surface there was slight dimpling. The growth and a small portion of the pedicle was removed. The patient recovered satisfactorily and remained well for five or six months, when she had similar symptoms of obstruction. Doctor Jones operated upon her again and found another melanoma causing intussusception about 12 inches above the ileocecal valve. The ileum contained several other growths, but there seemed to be no involvement of the liver. The patient's condition apparently did not permit resection of the tumor-bearing portion of the intestine. Six months after the operation, roentgenologic examination showed no metastases in the lungs. The patient died a few months later. There was no necropsy.

These cells obviously must have filtered through the lung and found a favorable soil in the submucosa of the small intestine, just as certain bacteria find a favorable tissue in which to grow.

MEGARECTUM AND MEGASIGMOID

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THE purpose of this paper is to record briefly two cases of segmental gigantism of the colon encountered in adults within a few months of each other, and to make certain comments on etiology and treatment.

CASE REPORTS

Case 1.—C. P., white, male, age 38, complained of obstinate constipation, recurrent fecal impaction and fecal incontinence. He had been born with an imperforate anus which was opened a day or so after birth by a stab-wound at the anal position. This

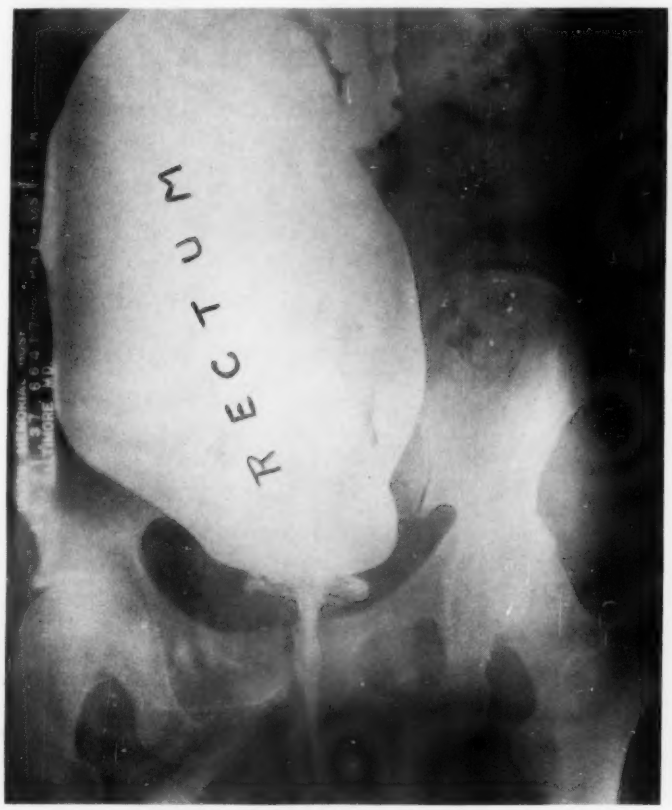

FIG. 1.—Case 1: Barium enema showing enormous rectum and sigmoid with normal descending colon.

proved inadequate as an outlet for the bowel, and on two or three occasions within the first few years of life, secondary operations were necessary to enlarge the anal opening. With this help, the patient, an unusually intelligent man, managed to grow up, get an education and fill the demands of a taxing occupation, but always under the handicap of great difficulty in defecation. He had practically no spontaneous desire for defecation

and absolutely no anal control. He had developed a technic of laboriously washing out the bowel about twice a week with repeated large enemata until he felt sufficiently relieved of the accumulated fecal mass. Between times, there was constant soiling from a little fecal seepage which necessitated the wearing of a pad continuously. Aside from this and recurrent attacks of abdominal distention and cramps associated with the impactions, his health was fairly good.

Physical Examination showed a young man of rather slender build and sallow color. His abdomen was flat above the umbilicus but with some fulness below it. The anal

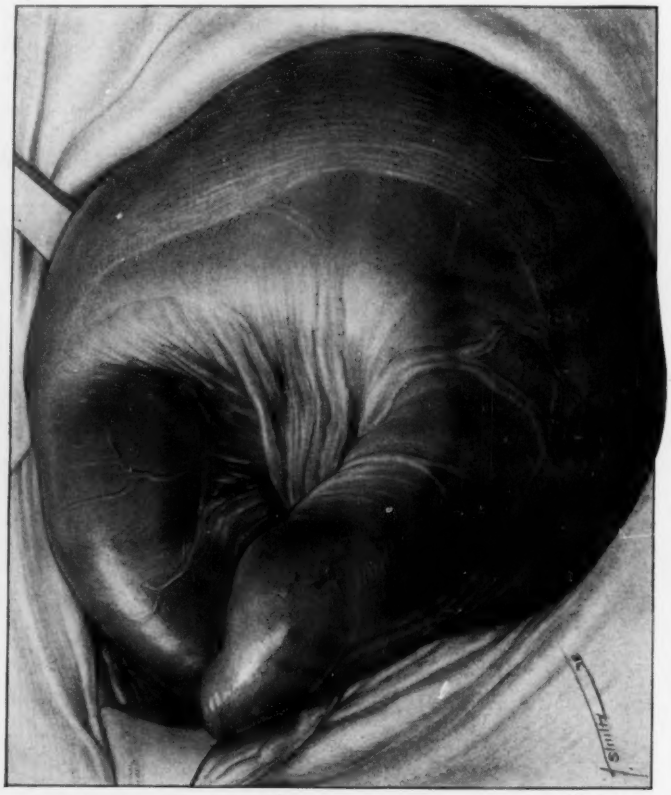

FIG. 2.—Case 1: Drawing showing the huge rectum and sigmoid.

orifice, in its proper anatomic position, was very small, about large enough to admit a No. 18 F. catheter, and rimmed about by dense scar tissue from frequent operations. The little finger tip could barely enter the rectum, so that digital exploration was impossible. A roentgenogram (Fig. 1) following a barium enema showed an enormously dilated rectum and sigmoid with apparently normal colon above. At the urgent desire of the patient, surgical relief of the condition was undertaken.

In considering the problem, three phases were apparent: The enormously dilated and redundant gut; the strictured outlet; and the absence of sphincter mechanism. It was decided, at a first operative attack, to remove the enormous fecal reservoir presented by the dilated rectum and sigmoid, and provide an adequate anal orifice. Later an attempt would be made to construct a sphincter apparatus. A week of preliminary preparation was carried out, with irrigations of the rectum with large quantities of fluid twice daily, the overnight instillation of oil, and restriction to a low residue diet.

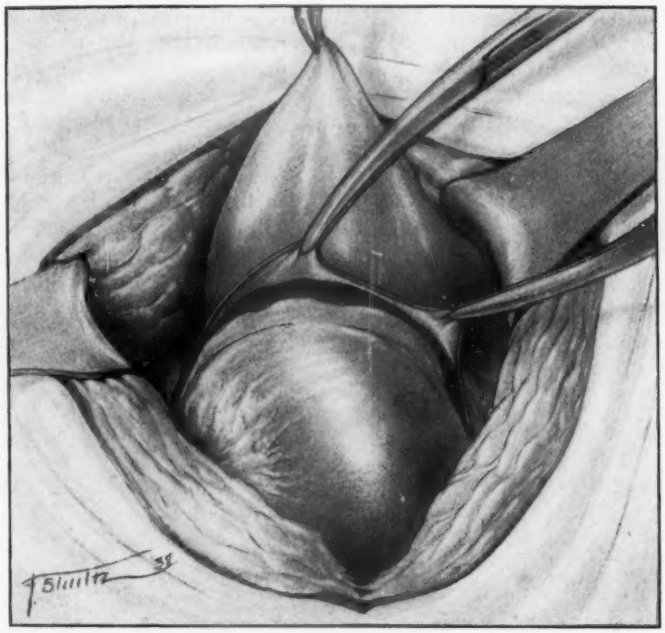

FIG. 3.—Case 1: Showing the division of peritoneum between rectum and bladder, mobilizing the rectum from above.

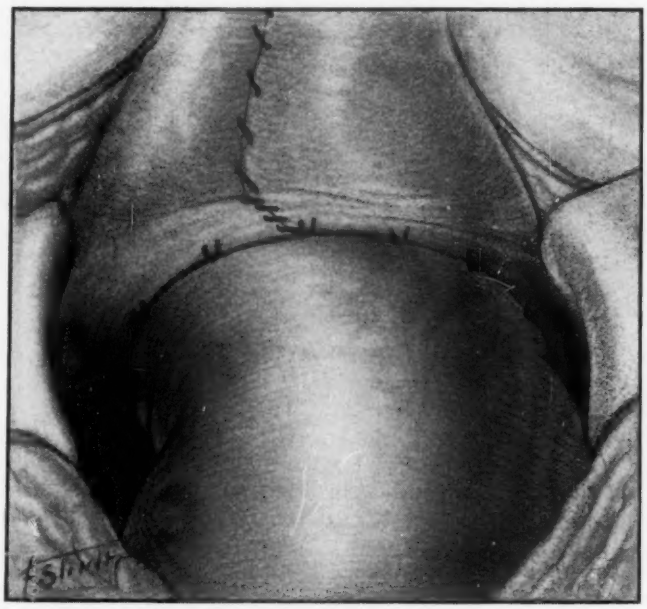

FIG. 4.—Case 1: Showing the repair of new peritoneal diaphragm after pushing down mobilized rectum.

First Operation.—Union Memorial Hospital, October 12, 1937: Under ether anesthesia, a midline incision, from symphysis to umbilicus, was made and a really astonishing rectum and sigmoid exposed (Fig. 2). The bowel from the upper two or three inches of sigmoid downward was greatly elongated, its walls thickened, and its diameter at least an average of four inches. In spite of efforts to clear the bowel beforehand, a fecal mass about the size of a grapefruit was found at the level of the sacral promontory. This was gently broken up by squeezing through the bowel wall and then milked downward into the lower rectum, but its presence was an annoying embarrassment throughout the operation. It was decided to proceed somewhat as in the first part of a Miles abdominoperineal resection for cancer, without, of course, ligating the inferior mesenteric vessels or transecting the gut. The peritoneum on both sides of the sigmoid mesentery was divided and also in front at the rectovesical reflection, so that the bowel was completely freed of its peritoneal attachment (Fig. 3). Mobilization was then continued by hand

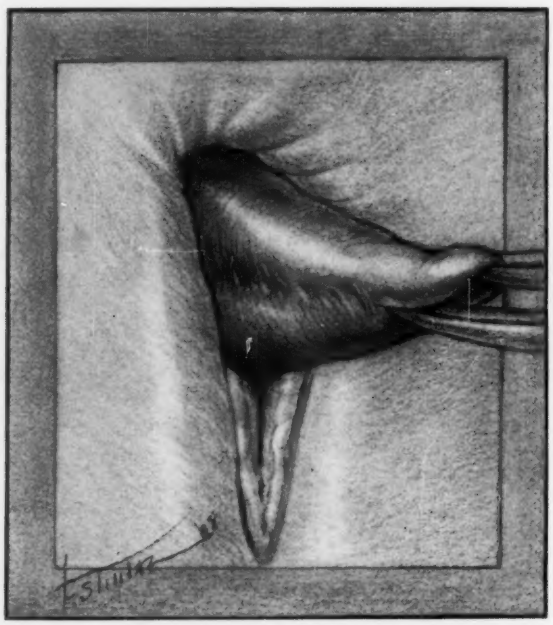

FIG. 5.—Case 1: Showing the incision about anus and beginning dissection of rectum from below, and the small, scarred anal opening.

dissection in the hollow of the sacrum down to the coccyx, and by division of the lateral and anterior fascial attachments. The mobilized bowel was then pushed into the pelvis and the pelvic peritoneum freed and closed over it to form a new pelvic floor (Fig. 4). Difficulty was experienced here because the great mass of gut and the fecal contents referred to above filled up the lower pelvis and prevented as complete an extraperitonealization of the abnormal bowel as was desired. The abdominal incision was then closed and the patient placed in the lithotomy position.

The scarred anal orifice was excised through a generous oval incision, which was then deepened upward and the rectal wall dissected from surrounding structures (Fig. 5). This dissection was continued until the area of mobilization previously accomplished from above was reached. The freed gut was now drawn out through the perineal incision as far as possible without undue tension and amputated (Fig. 6). The stump of bowel was united to the perineal skin by interrupted sutures, forming a new anus with a diameter

that would easily admit the index finger (Fig. 7). The resected bowel was about 16 inches long and from four to five inches in diameter.

In reconsidering this operation, an improvement suggests itself which will be used if a future opportunity presents. In the first stage, when the enormous gut has been thoroughly mobilized, it would be better not to reconstruct the pelvic floor. The abdominal incision would be loosely clipped together and securely draped; then the perineal stage carried out. This would allow the drawing down and resection of the bowel, unrestricted by the reconstructed pelvic peritoneal diaphragm. At the end of this step, the patient would again be put in the horizontal position, the clips removed from the abdominal incision, and the pelvic peritoneal diaphragm snugly formed about the sigmoid in its new position. It is believed that by avoiding this repair in the first part of the operation, the difficulty in crowding down the bulky gut into the lower pelvis would not arise, a more extensive resection could

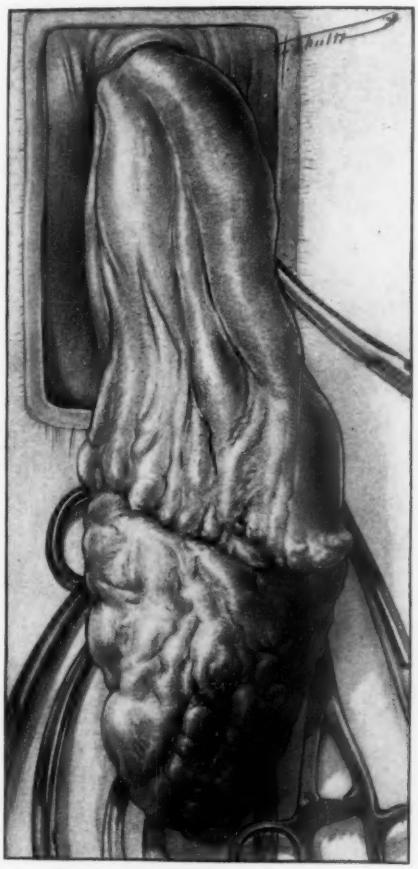

FIG. 6.—Case 1: Showing the giant rectum and sigmoid dissected from below and drawn out of perineal incision.

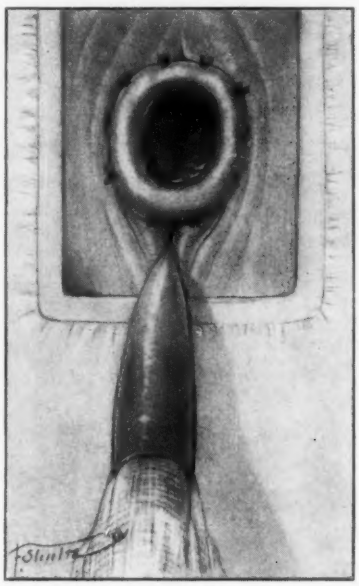

FIG. 7.—Showing the gut amputated; stump sutured to perineal skin.

be undertaken without this hampering feature, and a more secure pelvic diaphragm formed.

Recovery from this first operation was not particularly disturbed except by transitory urinary retention and the partial breakdown of the rectal suture line. This resulted in a moderate retraction upward of the rectal stump and healing by granulation. The patient was sent home for the completion of this process and general upbuilding before the second procedure. He was kept under observation and at the end of about three months showed a definite improvement in his nutrition and color, and he experienced a sense of well-being never previously enjoyed. The abdominal wound had healed *per primam*. The rectal wound was completely healed with a funnel-shaped, scarred outlet, narrowest

at its upper end, but which permitted the easy passage of the index finger. There was no anal control but the management of defecation had greatly improved. On only a few occasions, in several months, had impaction occurred, and it was easily removed by a simple enema. It was the patient's custom to take a mild saline laxative and this usually resulted in effectual evacuation. The amount of fecal soiling was much reduced. The bladder function was normal, but impotence had followed the operation.

Second Operation.—Union Memorial Hospital, March 15, 1938: Under ether anesthesia, a plastic procedure was undertaken, using two loops of fascia strips, one on each side, which were put in subcutaneously so as to encircle a bundle of the gluteus maximus muscle

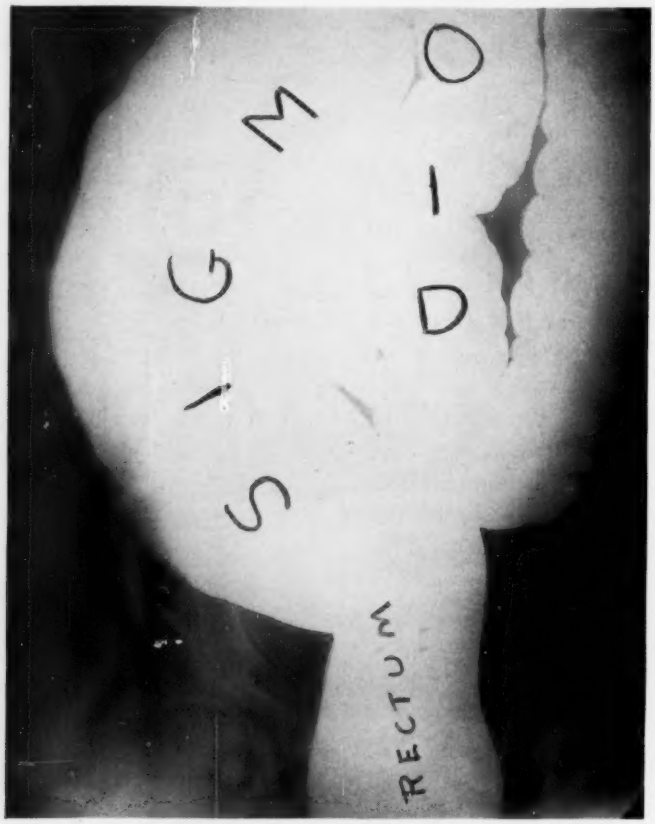

FIG. 8.—Case 2: Barium enema, showing giant rectum and sigmoid with comparatively normal descending colon.

at their outer ends and the anal canal at their inner ends. As this type of operation has been described elsewhere,[1, 2] it will not be reported in detail. There was a good deal of difficulty in executing it because of the extensive deep scarring about the anal orifice. The subcutaneous dissection to place the fascia strips and the compression of the anal canal on tightening them, were both interfered with by the scar tissue. At the end of the operation it was not felt to have been entirely satisfactory.

The patient again had urinary retention for a few days. The wounds became lightly infected and a part of the fascia was discharged. The final outcome of the operation was only indifferently satisfactory, since improvement of control was obtained, but far from what had been hoped for. The result of the whole treatment has been such as to please the patient by his definite improvement, but not to satisfy the surgeon.

Case 2.—E. L. P., white, male, age 71, had enjoyed excellent general health and digestion, including regular bowel movements daily without laxatives, until two years before admission to the Union Memorial Hospital, at which time, while traveling abroad, he became suddenly quite constipated. Laxatives were taken, without result. The condition became so serious that he entered a hospital, where by digital breaking up of an impaction and a number of enemata, the bowel was finally cleared out. A roentgenogram showed a huge rectum and sigmoid. Above the sigmoid the bowel seemed fairly normal. Roentgenologic examination at the Union Memorial Hospital confirmed this report (Fig. 8). Since the above occurrence, the patient has been unable to secure bowel movements without medication. By regular and careful attention to food and laxatives, with occasional enemata, he gets along fairly well.

Physical Examination reveals a man in generally good physical condition. The contour and size of the abdomen are natural and normal. There are no masses, fulness, tenderness or rigidity. On rectal examination, no stenosis, stricture or sphincter spasm can be felt. The anal orifice is of normal size and elasticity. No masses can be felt. The sigmoidoscope was passed with the greatest ease to its full length—11 inches—and the only abnormality noticed was the very large lumen of the bowel and slight congestion of the mucosa. *Clinical Diagnosis:* Giant rectum of unknown etiology. In view of the patient's age and the fact that medical measures sufficed to keep him symptom-free, no surgical treatment was advised.

Gigantism of the colon, from the time of Hirschsprung to the present, has attracted increasing attention. No attempt will be made to review the voluminous literature, as the cases herewith presented represent a special and small group under the general category of megacolon, namely, the limitation of the process to the rectum and sigmoid. The theory attributing megacolon to dysfunction of the sympathetic nervous system is not universally held, and in this group with limitation of the process to the rectum and sigmoid, David[3] has emphasized the part played by strictures and stenosis of the anal canal and low rectum. Warthen[4] has recently reported a case that seems to fall in this group. Case 1 is another such instance. One may say, therefore, that a number of cases of giant rectum and sigmoid occurring in early life are associated with anatomic obstruction of the outlet of the bowel. But the second case, herewith reported, cannot possibly be included in such a group. The patient reached the age of 69 with no evidence of trouble, and although his rectum and colon show much the same changes in size and loss of functional activity, there is not the slightest evidence of stricture, spasm or any other obstructive condition. One is forced to conclude that more than one etiologic factor may produce similar alterations in the lower segment of bowel. What the effective factor in Case 2 might be, one cannot do more than guess. Perhaps a slowly developing imbalance in a localized portion of the sympathetic innervation might explain it.

For obvious reasons, the methods of treatment employed in these two cases were entirely different. In the second case, a man of advanced years could be quite comfortable by suitable medical measures. In the first case, a young man, in the midst of an active career, was gravely handicapped by a condition due in large part to an anatomic disorder. Surgery seemed to offer the only chance of betterment, and although its results leave much to be desired, it has accomplished definite benefit. To those who may en-

counter such cases, the surgical method here employed, with the improvement suggested, may at least afford a point of departure for the attack on this problem.

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[4] Warthen, Harry J.: Multiple Congenital Abnormalities of the Colon. Southern Med. Jour., **31**, 528–529, May, 1938.

DISCUSSION.—DR. CARRINGTON WILLIAMS (Richmond, Va.): I would like to report the case of a boy, age nine, born with an imperforate anus. He had a perineal incision shortly after birth. During his whole life he has had attacks that amounted to complete intestinal obstruction from impaction of feces in the lower bowel. The abdomen was greatly distended when he was brought to the hospital, and it required strenuous treatment to empty the bowel. Roentgenologic examination disclosed a tremendous dilatation of the whole left side of the colon. I anticipated undertaking what Doctor Stone has described, but thought it safer to perform a colostomy. The anal canal was a rigid tube, about the size of one's little finger, and about the same length. I expected to bring the bowel through the skin and later to perform a fascial operation for control. After the colostomy had functioned for about ten days we examined the colon roentgenologically with a barium enema and, much to our surprise, the colon was found to be of normal size and the rigidity of the anal canal apparently had disappeared. We decided that any further operative procedure was unnecessary and we performed Doctor Stone's operation with strips of fascia for anal control before closing the colostomy stoma. We were fortunate enough to obtain good functional control, and a barium enema several months later showed a perfectly functioning colon. He has lost some control since that time, but there has been no contraction of the canal and no fecal impaction.

DR. HARVEY B. STONE (Baltimore, Md., in closing): Maybe I performed an unnecessary operation. I think there is a difference between a patient age nine and one age 38, however, and I question very much whether simple colostomy would have restored to normal this enlarged rectum and sigmoid in a man that age, although I think preliminary colostomy would have been a wise thing to try.

SURGICAL TREATMENT OF COMPLETE RECTAL PROLAPSE

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VARIOUS opinions have been expressed regarding the pathology, classification and particularly the surgical treatment of rectal prolapse. This is undoubtedly due to the fact that many varieties and degrees of rectal prolapse may exist. These vary from eversion of a small portion of the rectal mucous membrane to complete eversion of the entire rectum. A description of the operative procedures which have been used for the various degrees of prolapse would entail the writing of too long a paper; therefore, we shall confine our remarks chiefly to the presentation of an operative procedure which in our hands, at least, has been successful in the cure of complete rectal prolapse.

TYPES OF PROLAPSE.—"Prolapse of the rectum" and "procidentia recti" are terms with similar meaning and signify any condition in which part or the whole of the rectum is everted so as to protrude through the anal orifice. The condition has been classified by several authors as of first, second and third degree, or as partial and complete. Thus, the term "first degree prolapse" has been employed to denote a condition in which only the rectal mucous membrane protrudes; this condition is considered as a partial prolapse. "Second degree prolapse" and "complete prolapse" are terms used to express prolapse of all coats of the rectal wall. In this condition the prolapse may be of almost any extent and frequently is graded 1 to 4, depending on the extent of the protrusion; Grade 1 indicates minimal and Grade 4 maximal protrusion. The term "third degree prolapse" has been used to designate prolapse or intussusception of the colon into the rectum, without protrusion at the anus. This is really an intussusception of the rectosigmoid.

Etiology.—The cause of rectal prolapse is not clear but many interesting hypotheses have been presented. The rectum is held in position by several supports: The lower portion is supported by the pelvic fascia, the levator ani, the external sphincter muscles, the perineal fascia, the fibrous attachments to the coccyx and the prostatic or vaginal walls; the midportion, by the loose fibrous tissue which passes from the sacrum along the course of the lateral sacral arteries; and the superior portion, by the various peritoneal folds. For complete rectal prolapse to occur these supports must undergo some weakening or destruction, and some force capable of dislodging the organ from its position must be exerted. Occasionally congenital abnormalities, such as an extraordinarily long mesosigmoid and an abnormally deep cul-desac of Douglas or rectovesical pouch, as the case may be, are considered

to be important contributing factors. In general, if some anatomic weakness or abnormality is present, many exciting causes may initiate the prolapse. The condition is most often seen among individuals at the two extremes of life, infancy and old age, when general muscular tone is weak and patients have been subjected to a wasting disease of long standing, associated with some type of disturbance which causes excessive straining at stool.

Jeannel, in 1890, believed that prolapse of the rectum was due to a giving way of the upper attachments, while others were of the opinion that it was due to breaking down or loosening of the lower rectal supports. The etiologic view of most recent authors is that true rectal prolapse is actually a sliding, median perineal hernia through the pelvic fascia. This was first well explained and propounded in America by Moschcowitz,[2] in 1912. According to this view, the peritoneum at the bottom of the pelvic cul-de-sac is driven, by increased intra-abdominal pressure, downward through a natural defect in the pelvic fascia at the site where the rectum emerges from the abdominal cavity. Here the resistance offered by the perineal body prevents further downward progress of the hernia and its direction is thus diverted posteriorly onto the anterior wall of the rectum, which ultimately gives way to permit inward pouching of this wall. As the hernia proceeds, resistance is met posteriorly by the sacrum and coccyx and the course is again changed, at first in a downward and forward direction and finally backward toward the anus. According to Moschcowitz,[2] the prolapse in the beginning involves only the anterior rectal wall but later, as it enlarges, it draws in at first the two lateral walls and finally also the posterior wall, until further drawing in of the bowel is prevented by the firm fixation of the organ. It is not the purpose of this paper to attempt to prove that rectal prolapse cannot develop in the manner of a sliding hernia as propounded by Moschcowitz; however, it is our belief that an abnormally loosely attached rectum is the most important predisposing factor in its development and that prolapse cannot develop when the organ is firmly fixed.

In the presence of an abnormally loosely attached rectum, the result of a developmental defect, excessive straining at defecation over a long period, or perhaps some wasting disease, the force required to produce the eversion of the rectal walls through the anal canal is found in the propulsive movements of the rectum during the act of defecation, together with increased intra-abdominal pressure. A somewhat analogous condition occurs not infrequently at the site of colostomy when the bowel, usually the proximal limb, prolapses through the artificial stoma. This is especially likely to occur in those cases in which the segment of the colon which has been exteriorized has a long mesentery, and conversely, this complication usually does not occur if the colon just proximal to the site of colostomy is partially fixed by a short mesentery, as ordinarily prevails in the descending colon. While in the development of prolapse of the rectum there may be factors other than those at play in the production of prolapse of the colon through a colonic stoma, it is our belief that the general principles underlying the two conditions are similar.

Treatment.—In the main, five types of operations have been employed in the treatment of rectal prolapse: (1) That type which causes narrowing of the anus and rectum; (2) that which brings about restoration of the pelvic floor; (3) that which causes suspension or fixation of the prolapsed bowel; (4) that in which the prolapsed bowel is resected; and (5) that which is designed to obliterate the pelvic cul-de-sac.

The type of operation that should be employed will vary with the type and underlying causes of the prolapse present and with the age and general condition of the patient. All of the operations presented have been curative in a certain selected group of cases but it has been our experience that in each group there have been certain recurrences. Recurrence has been most marked among adult patients with complete rectal prolapse. The failure of many of the intra-abdominal operations employed to fix the rectum by sigmoidopexy can be explained by the fact that in some instances upward traction on the sigmoid, without first dividing the pelvic peritoneum along the line of its reflection at the sides of the lower part of the sigmoid, will not make the rectum taut. Therefore, since the operation fails to fix the rectum, benefit cannot possibly accrue from this procedure. Furthermore, in those cases in which the rectum can be made reasonably taut by traction on the sigmoid, without division of the pelvic peritoneum, recurrence is likely to follow because of the extreme difficulty in permanently fixing the sigmoid. The adhesions which bind the sigmoid to the pelvic and abdominal walls, promoted by the operative procedure of sigmoidopexy, will ultimately loosen because of the constant downward pull on the sigmoid and this, in turn, will permit the lower part of the sigmoid and the rectum to slide down along the hollow of the sacrum to occupy their original position. While the ostensible purpose of the Moschcowitz operation is to repair the defect in the pelvic fascia by obliterating the pelvic cul-de-sac, fixation of the sigmoid is an important feature of the operative procedure. The failures that result from this operation are due to the same cause: inadequate fixation of the rectum.

For many years it has been the practice of one of us (Pemberton), in order to aid in exteriorization of growths that are low in the sigmoid or in the rectosigmoid, to divide the pelvic peritoneum at its point of reflection and to free the rectum from the hollow of the sacrum down to the tip of the coccyx. Normally the posterior walls of the lower part of the sigmoid and of the rectum are only loosely attached to the hollow of the sacrum by areolar tissue. This portion of the bowel, then, can be pulled up out of the hollow of the sacrum and, in many instances, the lower part of the sigmoid and the rectosigmoid can be completely exteriorized as in a Mikulicz type of operation. This procedure leaves a cavity between the hollow of the sacrum and the rectum, and it has been our observation, after examination of the rectum in many of these cases, that this cavity filled in with a ridge of scar tissue which produced firm fixation of this portion of the bowel. It was reasoned that if the pelvic colon could be pulled up until the rectum was taut and held in this position, at least temporarily, by sigmoidopexy, the rectum would become

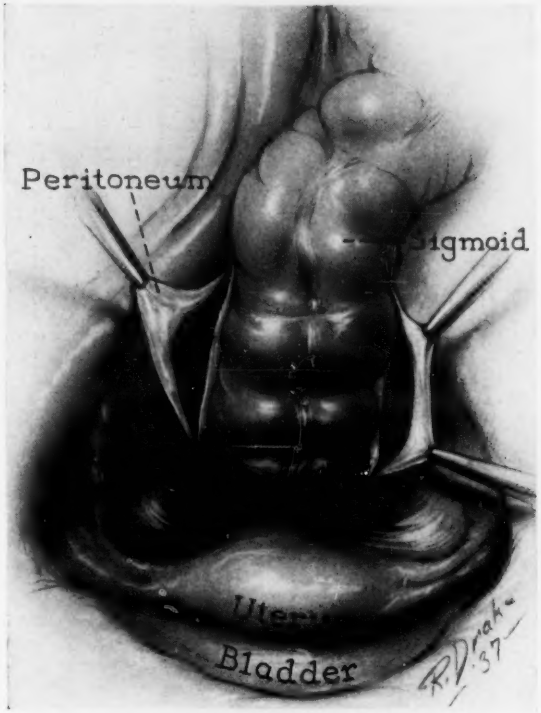

FIG. 1.—Incision of the lateral parietal peritoneum.

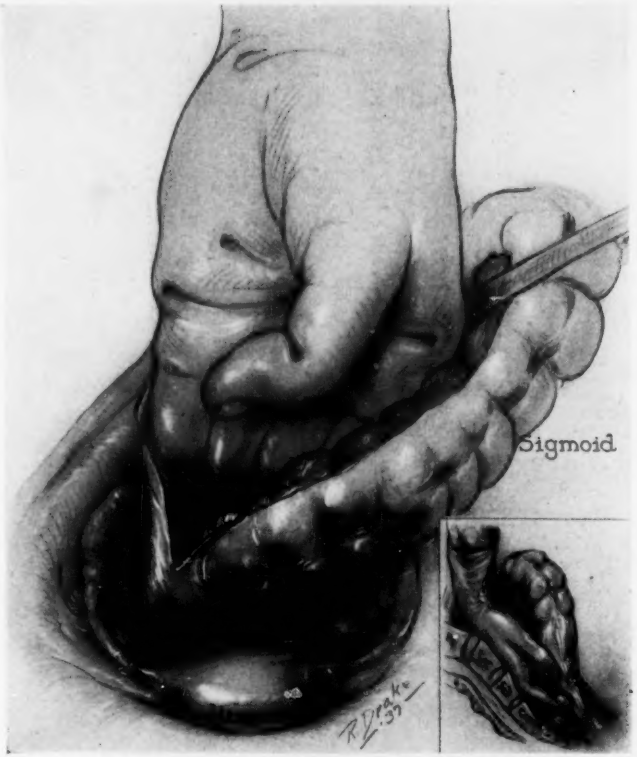

FIG. 2.—The sigmoid pulled upward and the rectum being freed from the hollow of the sacrum.

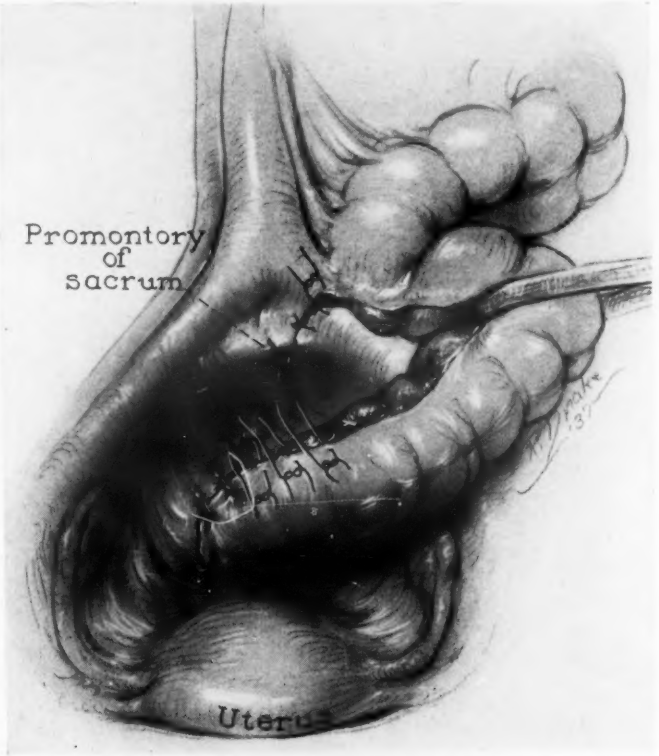

FIG. 3.—The prolapsed bowel elevated and reflected flaps of peritoneum approximated. The raw surfaces of the bowel are peritonized and the elevated bowel fixed in its elevated position.

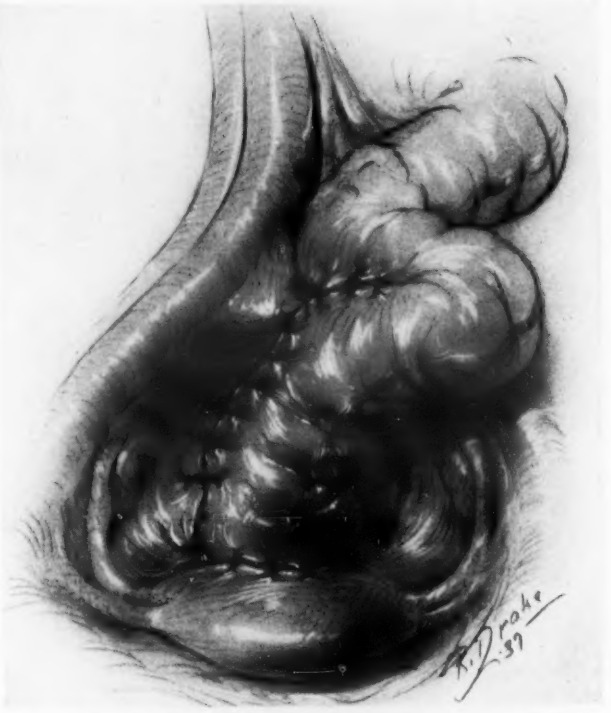

FIG. 4.—Fixation of the elevated bowel by attaching it to itself, to the uterus and to the walls of the pelvis.

fixed in this taut position by the formation of scar tissue in the hollow of the sacrum and complete rectal prolapse thereby could be cured.

Authors' Technic.—The patient is hospitalized for two or three days for preoperative preparation, during which time the bowel is emptied by administration of saline purgatives, colonic irrigations and use of a diet of which the residue is less than that of a regular diet but more than that of a nonresidue diet.

The operation usually is performed under spinal anesthesia. The patient is placed in the high Trendelenburg position and the abdomen is opened

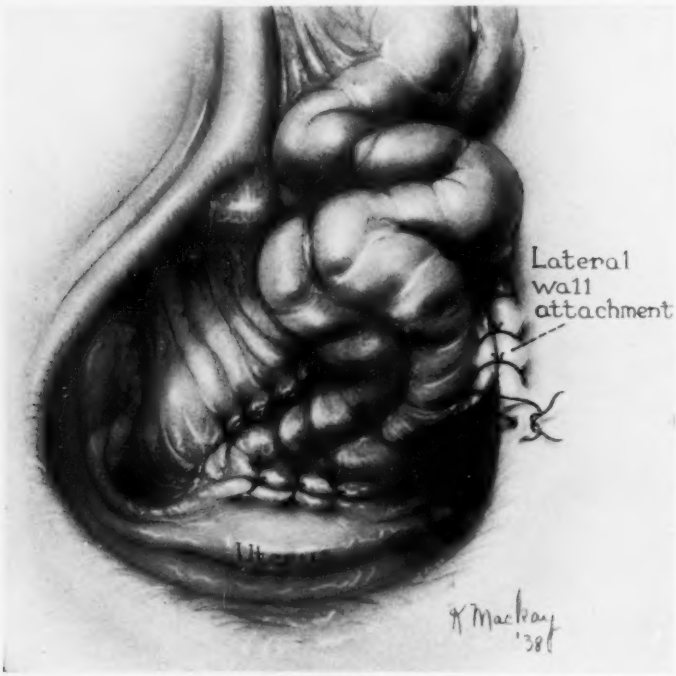

FIG. 5.—Fixation of sigmoid to lateral abdominal wall.

through a left paramedian incision which extends from the symphysis pubis to a level just above that of the umbilicus. A Balfour, self-retaining tractor is inserted, the small bowel and upper part of the abdomen are separated from the pelvis by a moist gauze pack and, if the patient is female, the uterus is retracted anteriorly.

The sigmoid, which is frequently found rather elongated and more mobile than usual, is gently pulled up until taut, and an incision is made in the peritoneum on both sides of the mesentery of the bowel and carried forward toward the bladder (Fig. 1). The peritoneum is not usually freed for a very great distance laterally, but it is usually advisable to identify the ureter on either side at the brim of the pelvis. Care also is taken not to injure the inferior mesenteric vessels and ligation of any of these branches is rarely necessary.

Next, the bowel is lifted and, with a hand behind it, in the hollow of the sacrum, the rectum is freed by blunt dissection (Fig. 2). The hand is carried downward and forward until the dissecting fingers are felt at the tip of the coccyx. This allows the prolapsed portion of the rectum to be pulled up from the hollow of the sacrum. With the rectum held taut, the reflected flaps of peritoneum are closed over this potential cavity and the raw surfaces of the bowel are peritonized (Fig. 3). It is important to fix this elevated portion of bowel in its raised position. This portion of bowel may be treated in various ways, depending on several factors: namely, its length and mobility and whether the patient is male or female. In the main, however, the bowel is fixed in its elevated position by suturing it to various portions of the abdominal wall and to the pelvic organs. Frequently it can be supported by

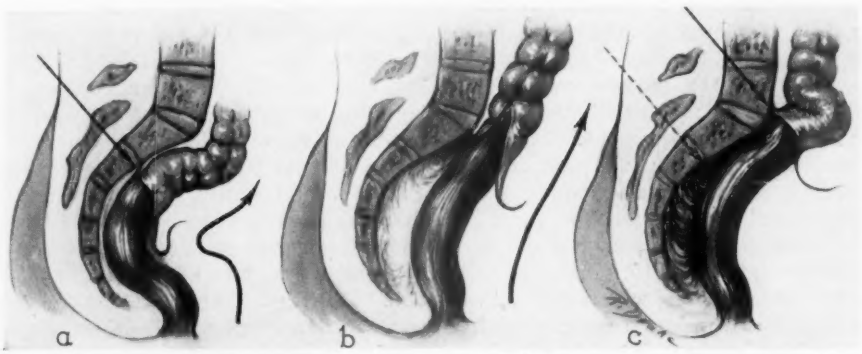

FIG. 6.—Diagrams of the operation: (a) The low level of the peritoneal reflection and the depth of the cul-de-sac of Douglas or rectovesical pouch. (b) The rectum freed from the hollow of the sacrum and elevated. (c) The peritoneal reflection and cul-de-sac of Douglas, or rectovesical pouch, have been raised and the elevated bowel has been fixed intra-abdominally. The cavity between the rectum and sacrum will fill with fibrous tissue.

suturing together a portion of the mesentery of the two loops of elevated bowel and further by attaching the bowel to the uterus (Fig. 4). In some cases the bowel is sutured to the peritoneum of the lateral walls and to the brim of the pelvis (Fig. 5); in other cases, to the anterior abdominal wall. In every instance care must be taken in placing the sutures onto the wall of the bowel lest leakage from the bowel results. Commonly the colon can be adequately anchored by suturing fat tags to the supporting tissues and, when it is necessary to suture the wall itself, the needle should pass through only the peritoneal coat. Care also must be taken not to leave an opening through which a portion of small bowel might herniate. Diagrams of the various steps in the operation are shown in Figure 6.

After the operation, in order to avoid gaseous tension in the rectum, a rubber tube is passed through the anus and is left in place for four or five days. During this period the bowels are kept constipated and a nonresidue diet is prescribed. Following this period the constipation is relieved by administration of oil enemata and of mineral oil by mouth. The diet is gradually increased and the stools are kept soft by frequent administration of oil. The

patient should remain in bed for two weeks following the operation and then should be cautioned to avoid strenuous efforts for three to six months.

During the past two years we have performed this type of operative procedure upon six adult patients with complete rectal prolapse; appended are brief reports of the cases.

CASE REPORTS

Case 1.—The patient, a male, age 44, was first examined at the Mayo Clinic, September 24, 1936. He had had a rectal prolapse since infancy and, in 1926, partial excision of the prolapsed organ had been performed elsewhere. There had been a recurrence after four years. At the time of his examination the bowel would gradually become prolapsed if the patient stood for any considerable length of time. There was some fecal incontinence and mucus discharge from the rectum.

The patient was thin, weighing 105 pounds (47.6 Kg.), and had very poor musculature. Proctoscopic examination revealed a relaxed, dilated anus; several small, sessile polypi in the lower part of the sigmoid and in the rectosigmoid, and complete rectal prolapse, Grade 4.

Repair for the prolapse was made, September 28, 1936, by the method described in this paper and, in addition, because of the depth of the rectovesical pouch and the marked degree of prolapse, partial obliteration of the true pelvis was effected. He was discharged on the twenty-first postoperative day. He returned after two months, at which time the polypi in the rectosigmoid were removed by fulguration. The rectum then seemed well fixed in normal position. There has been no recurrence.

Case 2.—The patient, a male, age 22, was first examined at the Mayo Clinic, December 30, 1936. He complained that for the past four years his rectum had been completely prolapsing with each bowel movement. This required digital replacement and was associated with pain and constant low backache. The symptoms and degree of prolapse were progressively increasing. The patient weighed 147 pounds (66.7 Kg.). Roentgenologic examination of the colon gave negative results but a rather marked angulation of the sacrum was revealed. Proctoscopic examination disclosed a small, pedunculated polyp on the right wall of the rectum at a distance of 14 cm. from the anus. This was removed by fulguration. On straining, the patient was able to produce complete prolapse of the rectum.

January 4, 1937: Intra-abdominal fixation of the rectum, as has been described, was performed. The patient was dismissed from the clinic on the eighteenth day after operation. There has been no recurrence.

Case 3.—The patient, a single woman, age 53, was first examined at the Mayo Clinic, August 26, 1937. She had been partially incapacitated for the past six years with almost constant rectal pain, which was most marked when she was on her feet. Frequently, when straining at stool, her rectum would become prolapsed to form a mass larger than her fist.

Physical examination revealed a rather psychopathic individual, weighing 125 lbs. (56.7 Kg.), but in good general condition. Proctoscopic examination revealed a relaxed anus, Grade 3, a polyp on the left wall of the rectum at a distance of 6 cm. from the anus and complete prolapse, which formed a mass 8 cm. long and 2.5 cm. in diameter.

August 26, 1937: Intra-abdominal fixation of the rectum was performed as has been described. The woman was dismissed on the nineteenth day after operation. There has been no recurrence.

Case 4.—The patient, a male, age 55, was first examined at the Mayo Clinic, February 15, 1938. Prolapse of the rectum had been present for 15 years. This had been aggravated during the course of pneumonia in 1936. A drugless healer had given several injection treatments for some associated hemorrhoids during June, 1937. Thereafter the rectal difficulty had been greatly aggravated. The prolapsing of the bowel, as well as its

reduction, was associated with considerable pain. Lower abdominal, cramp-like pain, associated with "gas" and occasionally with nausea, was brought on when the patient remained on his feet for any considerable time and was relieved when he lay down. Each bowel movement was associated with passage of varying amounts of bright red blood and, on two or three occasions, there had been a rather profuse hemorrhage from the rectum.

The patient weighed 180 lbs (81.6 Kg.), having lost 40 lbs. (18.1 Kg.) during the past year. Hemoglobin, 7.3 Gm. per 100 cc.; erythrocyte count, 3,660,000. Proctoscopic examination revealed localized proctitis, several large external hemorrhoids and a complete rectal prolapse, that could be produced by straining and that formed a mass measuring 8x8 cm. Roentgenologic examination of the colon gave negative results, as did all other examinations of the colon.

The patient was given a transfusion of blood and, on February 25, 1938, repair of the rectal prolapse was effected by the method that has been described. He was dismissed from the clinic on the 22nd day after operation. At that time the rectum was in normal position and there has been no recurrence.

Case 5.—The patient, a male, age 25, came to the Clinic, July 27, 1938, because of prolapse of the rectum. This had been of gradual onset, beginning 10 years previously; with each bowel movement the size of the prolapsed portion would increase. There had been no pain and no bleeding. In August, 1937, he had been operated upon elsewhere, at which time some redundant mucous membrane had been removed and a hemorrhoidectomy performed. Following the operation there had been some improvement, but shortly before the patient's visit to the clinic the prolapse had become progressively worse.

Examination revealed that a prolapse was produced on straining at stool. All coats of the bowel were involved and the prolapsed portion was about the size of an orange.

August 1, 1938: An internal fixation operation, according to the method that has been described, was performed. The patient's convalescence was uneventful and he was dismissed from the Clinic, August 23, 1938. The rectum was in normal position at that time.

Case 6.—The patient, a female, age 54, registered at the Clinic, November 2, 1938. She complained of "hemorrhoids" of three years' duration and of prolapse of the rectum when on her feet. She was able to reduce the prolapse by deep inspiration. Occasionally, when she was on her feet and the prolapse was present, her bowels would move involuntarily. She had noticed bleeding from the rectum only once.

The woman was obese and had a cystocele and a rectocele. Laboratory findings were essentially negative. Roentgenologic examination revealed that the gallbladder was functioning poorly. Proctoscopic examination disclosed rectal prolapse; the prolapsed portion measured 6x6x5 cm.; the anus was patulous.

November 7, 1938: An intra-abdominal fixation operation was performed for repair of the rectal prolapse. The rectum was loosened from the hollow of the sacrum and traction was made on the lower part of the sigmoid so as to lengthen the distal limb of bowel. After the openings in the peritoneum had been resutured, the rectum and sigmoid were sutured to the right lateral pelvic and abdominal walls and also to the posterior surface of the uterus, which was found prolapsed with the prolapse of the rectum.

The patient made an uneventful recovery and was dismissed from the Clinic, November 28, 1938. Her general condition was excellent and the rectum was in normal position.

COMMENT

We realize that the success or failure of an operation cannot properly be evaluated until a large group of patients have been studied over a long period. Neither have we had a large group of patients, nor have we had the opportunity to study our postoperative results over a long period. However, in view

of the many poor results that have been obtained in advanced cases of complete rectal prolapse when other operations have been employed, and because of the theoretic soundness of this operation, and the excellent immediate results that we have obtained with it in six cases, we were encouraged to present it for consideration.

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PANCREATIC CALCULI

A REVIEW OF SIXTY-FIVE OPERATIVE AND ONE HUNDRED THIRTY-NINE NON-OPERATIVE CASES

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PANCREATIC calculi were first observed and recorded by Graaf, in 1667. Later, Morgagni (1765) and Cawley (1788) reported autopsy findings of stones.

Capparelli (1883) is given credit for the first operation for the removal of pancreatic calculi. He records the case of a baroness who developed an abscess above the umbilicus after many attacks of epigastric colic. The abscess ruptured and discharged pus and gritty material. The resulting pancreatic fistula persisted for six years and over 100 calculi were discharged. The fistula then closed spontaneously, but diabetes developed and death soon followed.

Mr. Pearce Gould, of England, in 1891, eight years later, successfully removed several stones from the duct of Wirsung, but at a second operation 17 days later found and removed a large stone near the duodenum, which was obstructing both the duct of Wirsung and the common duct. The patient, however, died of "exhaustion," 12 days after the second operation.

In 1902, Moynihan diagnosed and successfully removed a pancreatic stone through the duodenum. The symptoms which he described in this first successful case of stone in the duct without abscess were as follows: "Steady loss of health, gradual wasting, irregular pigmentation in the skin in patches of the color of café-au-lait, they very closely resembling the pigmentation of molluscum fibrosum, persisting attacks of epigastric pain and uneasiness of a type of hepatic colic no less severe, and unattended, until very late in the history, by jaundice, which was then always trivial though unmistakable, and pain passing through from the front of the abdomen to the middle of the back. There was no rigor or any complaint of sensations of heat or cold. Stools were occasionally frothy and 'greasy.' On examination, under chloroform, some indefinite swelling could be felt above the umbilicus and a little to both sides of the median line, though chiefly to the right."

In 1925, Seeger[62] collected 23 cases operated upon for pancreatic stone and 80 cases in which pancreatic stone had been found at autopsy. Since his report in 1925, we have collected 42 additional operated cases, including one of our own making a total of 65 operative cases in 55 years. More than half of them have occurred during the past 14 years. Of these 65 cases, stones were removed at operation in 58, with an operative mortality of 17.2 per cent (ten deaths.) The mortality of the entire 65 cases was 18.4 per cent (Table I).

Since the 80 autopsy cases tabulated by Seeger in 1925, we have found 30

TABLE I

RÉSUMÉ OF 65 OPERATED CASES OF PANCREATIC CALCULI

Reported	Age Sex	Pain	Other Symptoms	Jaundice	Gallstones	Glycosuria	Pathology	Outcome
Caparelli, 1876						Sugar late in disease	Many stones in an abscess	Recovered. Death years later from diabetes
Prewitt, 1898			Weight loss, tumor				One stone in abscess	Not stated
Gould, 1898	46 M.	Absent	Loss of weight, strength and appetite. Clay-colored stools	Marked	None	None	Pancreas and liver enlarged. Pancreas contained several stones in body and in duct of Wirsung. Stones were calcium carbonate	Died, twelfth day. Peritoneal cavity clean except for abscess in region of cecum
Dalziel, 1902				None	None		One stone in duct of Wirsung, size of a pea	Recovery
Moynihan, 1902	57 F.	Epigastric, type of hepatic colic but less severe. Radiated to back	Wasting of flesh. General health loss. Pigmentation of skin in patches. Greasy, frothy stools. No chills or fever	None	None	None	Pancreas enlarged, especially head. One stone in terminal portion of duct	Recovery
Allen, 1903 (Clark-Burnam)	30 F.	Paroxysmal, epigastric radiating to left lumbar area	Lost 50 lbs. in 1 yr. Chills and fever. Clay-colored stools	Early for 1 wk. Not with later pain	None	None	Two cysts of pancreas containing 2 stones. Calcareous material throughout pancreas, which was atrophic. Autopsy report showed islands of Langerhans preserved	Died, fifth day. Autopsy showed no peritonitis
Mayo Robson, 1904	57 F.	Severe			None	None	One stone in duct of Santorini, 2 in duct of Wirsung, one in pancreas	Recovery
Hollander, 1905	48 M.	None	Marked jaundice; emaciation. Enlarged liver and spleen. Epigastric tumor (late); clay-colored, fatty stools	Marked	None	None	Pancreatic cyst containing fluid containing cholesterol and one calcium carbonate stone	Recovery
Staehlin and Roeber, 1905	39 F.	Deep seated, boring pain in epigastrium. Intermittent attacks	Fatty stools	None	None	None	Four stones in head of pancreas	Recovery

Ruth, 1907	27 F.	Upper abdominal colic	Long history. Never strong. Stools normal	None	None	None	Pancreas large. Many calcium carbonate stones in duct in body of pancreas. Some were polished by movement of aorta	Recovery
Link, 1911	22 F.	Paroxysmal, left kidney region, radiating down ureter	Emaciation; anemia; intermittent hydronephrosis suspected and operation for this performed. Kidney normal. Stools normal	None	None	None	Multiple calculi with marked interstitial pancreatitis	Recovery
Friedlander, 1911	54 F.	In attacks. Upper right abdomen	Fever and vomiting with attacks	Following attacks	Present	None	Cholesterol stones in gallbladder, common duct, pancreatic duct, and head of pancreas. Chronic cholecystitis	Recovery
Murray, 1911	40 F.	Severe, epigastric in attacks	Long history. Operation for gallstones. None found. Six wks. later operation for pancreatic stone	With later attacks	None	None	Gallstones. One stone in pancreatic duct	Recovery
Lacoutre and Charbonnel, 1914	32 F.	Violent attacks, upper right abdomen radiating to back	Vomiting	None	None	None	Two calcium carbonate stones, one in head of pancreas and one in wall of gallbladder	Recovery. Later X-ray showed overlooked stone
Dowd, 1915	32 F.	Severe epigastric, referred to pancreas	History over several years. Passed mulberry calculi in stools	None	None	None	Fat necrosis. Abscess in head of pancreas. 30 or more stones in the abscess	Recovery
Hartig, 1915	34 M.	Upper abdominal radiating to shoulders	X-ray negative; stools normal	None	None	None	Two calculi in a carcinoma of head of pancreas	Recovery for 5 mos. Death due to carcinoma
Navarro, 1921	? F.	Pain similar to that of gallstones	Dull ache over pancreas	Recurring	Present	None	Three calculi in pancreas. Cholecystitis with stones	Recovery
Bost, 1921	26 M.	Violent epigastric cramps radiating to back and along left costal margin	15 yrs. duration. X-ray and gastric analysis negative. Epigastric tenderness and rigidity. Nausea and vomiting. Temperature 103°–104° F. Leukocyte count 26,000. Underweight	None	None	None	Head of pancreas much enlarged, 3 to 4 times normal size. Number of calcium carbonate stones removed. Also thick pus drained. (Biliary tract normal)	Recovery. (Had pancreatic fistula for 6 wks.)
Sistrunk, 1921	48 F.	Epigastric, cramp-like	Diarrhea. Loss of weight and strength	Marked			Had a duodenal ulcer. Gastroenterostomy made. Spleen large. One stone about 1½ ins. above ampulla in pocket of pancreas	Recovery

TABLE I (*Continued*)

Reported	Age Sex	Pain	Other Symptoms	Jaundice	Gallstones	Glycosuria	Pathology	Outcome
Sistrunk, 1921	33 M.	Colicky, several years	Operated upon 1 yr. before for gallstones. Pancreas found to contain stones then, but these were not removed until later operation		Present	Present	Many stones in duct of Wirsung and pancreas. Some in duct of Sanatorini (not removed). Appendix diseased	Increased sugar tolerance. Recovery
Sistrunk, 1921	68 F.	Epigastric, radiating to left	15 lb. weight loss				Many stones in head of pancreas, apparently in main duct	Recovery
Sistrunk, 1921	62 F.		Duodenal ulcer history 15 yrs.				Ulcer found. Gastro-enterostomy. One stone in pancreatic duct near ampulla	Recovery
Lindsay, 1922	29 M.	Violent epigastric radiating to back and to left	15 yr. history. X-ray showed shadows outside stomach. Stools normal. Irregular pigmentation on face, neck, and chest. Slight wasting for 2 yrs.	None	None	None	Six large and many small stones of calcium carbonate in pancreas. The gland tissue was thinned and the ducts dilated and thickened. Cultures sterile	Recovery
Jarboe, 1922	56 F.	Colicky, 3 yrs.	Weight loss	None	Present	Present	Number of stones removed	Recovery
Jarboe, 1922	47 F.	Mild	Indigestion	None	Present	None	Stones not removed	Recovery
Hartman, 1925	20 F.	Periodic attacks, increasingly severe, high in the epigastrium; radiation to the midscapular region	Vomiting, leukocytosis 19,600; no fever. No weight loss	None	None	None	Elongated tumor mass, 6 cm. in diameter, made up of pancreatic tissue and ducts; filled with many pancreatic stones, large—4 Mm. in diameter, smallest like sand. Small cyst at one point which also contained stones; tumor attached to head of pancreas	Tumor removed. X-ray taken after operation, showed multiple shadows in right side opposite first lumbar vertebra, believed to be pancreatic calculi
Hartman, 1925	33 F.	Dull soreness in epigastrium, worse after meals	No weight loss. No other symptoms. Pelvic tumor present	None	None. Moderately severe chronic catarrhal cholecystitis	None	Many stones in head of pancreas	Stones which were easily accessible removed. Immediate postoperative convalescence normal

Hartman, 1925	45 M.	Dull aching in epigastrium immediately after meals. Pain never severe	No weight loss. Vomited "coffee grounds" short time before operation. Epigastric tenderness. Duodenal deformity seen by X-ray	None	None	None	Marked hepatitis, probably cause of gastric bleeding; marked catarrhal cholecystitis; calcified irregular mass 2.5 cm. in diameter in contact with tail of pancreas, believed to be pancreatic in origin	Uneventful convalescence
Hartman, 1925	45 M.	Classical history of duodenal ulcer, 8 yrs. duration	No weight loss. High gastric acidity. X-ray showed duodenal ulcer	None	None	None	Stone 1 cm. in diameter, lower edge of body of pancreas. Duodenal ulcer	Uneventful recovery following gastro-enterostomy. The pancreatic stone probably caused no symptoms
Seeger, 1925	42 M.	Mild epigastric	Indigestion. Vomiting	None	None	None	Irregular stone removed	Recovery
Mraz, 1926	42 M.	Severe epigastric 5 mos. duration	Nausea with attacks	None	None	None	45 stones removed from head	Recovery
Perman, 1926	66 F.	Left epigastric pain severe	X-ray diagnosis of gallstones	None	Present	Present	Three pancreatic stones. Autopsy later revealed carcinoma	Died, 3 mos. after operation
Perman, 1926	67 M.	Severe epigastric	Grayish stools. X-ray diagnosis pancreatic stones	Present	None	None	Stones removed first operation. Common duct dilated second operation	Died. 3 wks. after operation
Dangis and Soschin, 1926	43 M.	Moderate	Cholecystostomy 3 mos. Recurring biliary fistula	None	None	None	Cholecystectomy. Autopsy showed pancreatic stones	Died, fifth postoperative day
Friedrich, 1927	24 F.	Colicky epigastric	Vomiting	None	None		Cyst of pancreas 6 liters in size with several stones	Recovery
Orth, 1927	46 M.	Severe; 10 yrs.		Present	None		Large calcareous mass excised from body	Recovery
Quenu, 1927	45 M.	Severe; 15 yrs.	"Hepatic colic"	None	None		Postoperative diagnosis carcinoma. X-ray showed pancreatic stones	Recovery
Gross, 1928	67 M.	Mild; 22 yrs.	Sense of pressure	None	None		Yellow stone from duct of Wirsung	Recovery
Wolf and Tietze, 1928	45 M.	Moderate	Diagnosed preoperatively	None	None	None	Ten stones removed	Recovery
Lindsay, 1928	43 M.	Radiation to left shoulder	Weakness. Polyuria	None	None	Present	Several stones and "sand" removed. Fibrosis of pancreas	Recovery
Calzavasa, 1929		Mild	Diagnosed by X-ray	None	None	None	Four large stones removed	Recovery
Zukockwerdt, 1929	46 M.	Colicky	Vomiting	Present	None		No stones removed	Recovery

TABLE I (*Continued*)

Reported	Age Sex	Pain	Other Symptoms	Jaundice	Gallstones	Glycosuria	Pathology	Outcome
Piquant, 1929	23 M.	Intermittent and radiating to back	Marked weight loss	Present	None	None	Stone size of hazelnut. Pancreatic fistula 2 mos.	Recovery
Irsigler, 1931	37 M.	Severe; 2 yrs.		None	None		Bloody fluid. Four stones found at autopsy	Died, fifth postoperative day
Stropeni, L., 1931		No data					Stones removed from main duct and pancreas	Recovery
Bost, 1931	36 M.	Cramps upper left side and back, also over left kidney and referred to left shoulder	Attack 10 days duration. Nauseated. Slight tenderness upper left side. No rigidity. Stools and previous gastric analysis negative. X-ray: multiple stone shadows in head and tail of pancreas	None	None	None	Adhesions from previous condition. Stones palpated in head of pancreas. Large mass about tail of pancreas. Number of stones and thick pus with colon odor drained from mass	Recovery
Vsland, Olav, 1932	36 M.	Severe pain right costal margin	Nausea. Temperature 38° C.	Present	None	None	Four stones removed from swollen pancreas. Fistula for 25 days	Recovery
Billaudet, 1933	58 F.	Mild	"Hepatic colic"	None	Present		Large, irregular calculus	Recovery
Soupault, 1933	M.	Referred to back	Weight loss	Present			(1) Cyst drained. (2) Two stones in fistulous tract. (3) Removal 4 stones through ampulla of Vater	Died, 36 hrs. after third operation
Thomason, 1933	14 M.	Moderate	Vomiting and weight loss. Diagnosis: "Appendicitis"	None	None	None	(1) Appendicectomy. Pancreas crepitant. (2) Small cyst with "sand"	Recovery
Collins, 1934	73 F.	Severe; many years	Indigestion	None	Present	None	One pancreatic stone	Died, third postoperative day
Rienhoff and Lewis, 1934	33 M. Col.	Acute pancreatitis, severe epigastric	First operation	None	None	None	Single stone removed second operation	Recovery
Tzschirntsch, 1935	19 F.	Moderate; 3 yrs.	Dysmenorrhea	None	None		Stone palpated but not removed	Recovery
Faust, 1935	45 M.	Moderate; 10 yrs.	Indigestion	None	None		Two-stage drainage. Stone removed at autopsy	Died, sixth postoperative day

Faust, 1935	45 M.	Moderate; 10 yrs.	Indigestion	None	None		Two-stage drainage. Stone removed at autopsy	Died, sixth postoperative day
Mayo, J. G., and Walters, 1935							16 stones removed through separate incision	Recovery
Carnot, 1936	F.	Mild	Indefinite indigestion	None	Present	None	One stone removed, many not removed	Recovery
Labbe, 1936	32 M.	Severe; 7 yrs.	Loss of weight	Present	None	None	Olivary stone removed	Died
Picot, 1936	30 M.	"Hepatic colic"	Emaciation	Present	None	Present	Large single stone obstructing duct of Wirsung	Died, eighth postoperative day
Carteret and Folliasson, 1936	45 M.	"Left subphrenic abscess syndrome"	Hepatic colic 20 yrs. before	Present	None	Present	(1) Drainage abscess tail pancreas. (2) Cholecystostomy	Died, fifteenth postoperative day
Bergeret, 1937	43 F.	Severe	Emaciation	None	None	None	Many stones in different compartments	Recovery
Herman and Guis, 1937	48 M.	Mild	Hyperinsulinism	None	None	None	Calcareous tumor of head	Recovery
Witherspoon, 1937	54 M.	Severe; 2 mos.	Loss of weight. Diagnosed by X-ray	None	None	None	Stones not removed. Cirrhosis of liver	Died within several days
Papin, 1938	54 M.	Severe; 10 yrs.	Loss of weight. Indigestion	Present	None	None	Two groups of stones in pancreas	Recovery
Kini, 1938	39 M.	Moderate; 8 yrs.	Indigestion	None	None	None	Three stones removed	Died, third postoperative day
Haggard and Kirtley, 1938	35 F.	Severe attacks; 24 yrs.	Previous cholecystostomy	Present	None	None	Several masses of stones from duct. Fistula 5 + mos.	Recovery

additional autopsy cases in the literature. There were also 28 cases in which a positive roentgenologic diagnosis has been made, and for various reasons were not operated upon.

Loeper and Bioy[42] report the unusual case of a young man with glycosuria, who found an irregular stone, not shown roentgenologically, in his stool, 24 hours after a crisis of pain, and has had no further attacks, and his urine has remained sugar-free. Those additional cases bring the nonoperative cases to 139 and all cases of pancreatic stone, both operative and nonoperative, to 204.

TABLE II

TOTAL CASES OF PANCREATIC CALCULI FOUND IN THE LITERATURE DURING 271 YEARS

Year	Operative Cases	Stones Removed at Operation	Mortality Percentage	Autopsy Cases	X-Ray Cases Not Operated
1667–1925 (Seeger)	23	22	8.5%	80	
1925–1938	42	36	25.0%	30	29
Totals	65	58	17.2%	110	29

Nonoperative cases 139
Total of all cases 204

The fact that the number of reported cases of pancreatic calculi has increased from 102, in 1925, to 204, in 1938 (Table II), would indicate that this condition is not so rare as is generally believed. The increased number of upper abdominal roentgenograms on patients having attacks of abdominal pain are adding to the recorded cases as well as increasing the accuracy of preoperative diagnoses. In 1925, less than four cases had been correctly diagnosed before operation. Since then a great many cases have been diagnosed either before operation or at autopsy and in patients not consenting to operation. Opie found the incidence of pancreatic stones to be one in 1,500 autopsies. Zeckwer[79] reported five cases in 10,300 hospital autopsies. They are more frequent in diabetics, three cases in 300 autopsied diabetics being reported by Warren.

Etiology.—The cause of pancreatic stones is not known. Stones in the pancreas may be divided into true pancreatic stones, which are found in the duct, and false stones, which are calcifications in the parenchyma of the pancreas, probably resulting from pancreatitis. In the latter, the calcification is in all probability due to pancreatitis secondary to disease of the gallbladder. True stones may result from infection, with stasis and altered pancreatic secretion being factors in their production.

It was recognized, before 1900, that pancreatic stones were mainly composed of calcium carbonate, which is not a constituent of pancreatic secretion, so the secretion must have become altered, due to stasis or infection. Pancreatic stones have been produced by the injection of sterile soot into the main duct (Thiroloix). Due to the large amount of carbon salts in these stones, they are clearly visible on a plain roentgenogram, and this is the surest and the only reliable diagnostic procedure.

The stones vary in size from a small pea up to a walnut. They tend to be multiple and often fuse to form branching, irregular masses.

Stones have been found in both the ducts of Wirsung and of Santorini and in dilated branches of these ducts. They are also commonly found in cysts and ectatic cavities in the parenchyma. They vary in number from one to 300.

In an autopsy case reported by Seres[64] (1934) the cut surface of one of the pea-sized stones found in the main duct showed 34 different strata when stained with Mallory's aniline blue. Part of them were composed of fibrin with concentric granules of calcium. Some of the granules contained gram-positive Cocci, Diplococci, and short and long chain Streptococci.

Pancreatic stones have a whitish-gray cast and are quite hard. They resemble salivary calculi in composition and appearance. They are more opaque than gallstones and stand out sharply on comparison in a roentgenogram. Kirklin,[63] referring to the roentgenographic evidence in two of the Mayo Clinic cases, spoke of a case where multiple pancreatic calculi were found in a tumor 5x7 cm. attached to the head of the pancreas, which the pathologist reported as an accessory pancreas in a girl, age 20, with weakness and palpitation for two years. She had, while under observation, high epigastric pain with vomiting.

Chase[63] discussed five cases diagnosed by him roentgenologically. One showed stones not in the gallbladder, during a Graham test, another lying in the curve of the duodenum, following a barium meal. The patient was a diabetic with epigastric pain which radiated through to the back.

Attention was called to the differentiation from calcareous deposits in the abdominal aorta, where the calcareous flecks lie in a line parallel with the spine. Also from calcareous mesenteric nodes where there was a mulberry-like shadow, and others are likely to be seen throughout the abdominal shadow.

Since ligation of the pancreatic duct does not cause retention cysts, it is questionable if impacted stones do. The process is probably gradual and similar to the hydronephrosis seen with partial, gradual obstruction. Certainly stones are associated with cysts in many cases and also with abscesses, intralobular fibrosis, and carcinoma (Fig. 2). The marked dilatation of the ducts in most cases indicates gradual obstruction. Friedrich and Hoesch[26] report an unusual case of a huge cyst of the pancreas, which contained six liters of mucoid liquid, and the preoperative roentgenogram had shown stones outside the gallbladder. Following drainage of the cyst, the stone shadows disappeared between the third and fifth weeks, presumably being passed into the bowel.

Bosq[8] reported three groups of calculi in a bull in addition to two cases in man. There were also small stones lying just beneath the capsule of the pancreas. Walters and the late Jos. G. Mayo reported a case in which stones were removed through separate incisions.

Symptomatology.—Pain is the only symptom present in every case of pancreatic stone which was operated upon. In the majority of cases the pain

was colicky and severe in character and radiated to the left side of the back. However, the pain so often resembles gallstone colic that the localization and radiation of the pain usually leads to the erroneous diagnosis of cholelithiasis. The term "hepatic colic" is widely used on the continent in describing the pain of pancreatic origin.

Localization of pain and residual tenderness are more often in the mid-epigastric region than with cholecystitis. The interval between attacks in

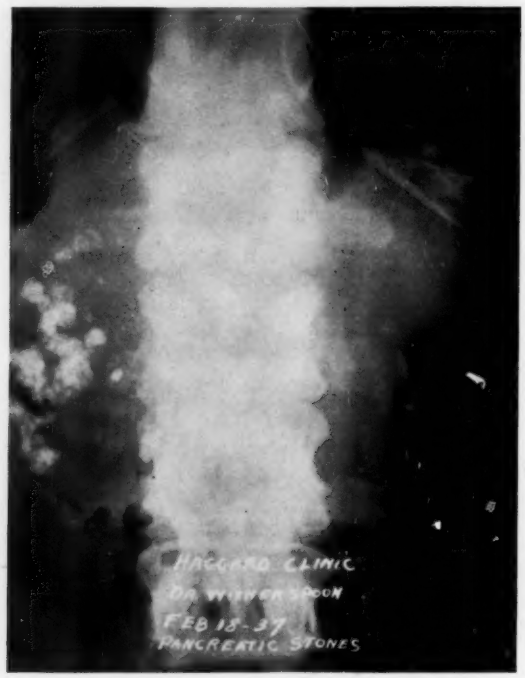

FIG. 1.—Pancreatic stones (Doctor Witherspoon's case). Exploration by Dr. W. C. Dixon showed carcinoma at head of pancreas.

pancreatic stones is longer than that in gallstones, and in many cases the attacks extend back to adolescence.

Vomiting is the next commonest symptom and is frequently described as bilious vomiting with increased salivary flow. Weight loss and even emaciation are present in about 75 per cent of the operative cases and even more frequently in the cases coming to autopsy. This is due to the large number of diabetics who also have pancreatic calculi, though glycosuria was rarely reported in this series.

Jaundice was present in 18 of the 65 operative cases studied (27.6 per cent) and tends to recur with each attack. In our case, jaundice was present in ten of the 12 attacks of colic. It is mild in most cases and often transitory.

Associated gallstones were present in nine cases (13.8 per cent), which argues against initial disease of the biliary tract as being the main case of pancreatic stones.

Glycosuria is reported in only six of the 65 cases (9.2 per cent). It is especially likely to occur with attacks, being absent in the intervals. Glycosuria is more common in the cases of long standing where considerable fibrosis of the gland has taken place. As pointed out by Moses Barron, in 1920, extensive destruction of the acini may occur without much damage to the islands of Langerhans, but in many of the autopsied cases even very little islet tissue could be found and there was marked atrophy of the gland. While

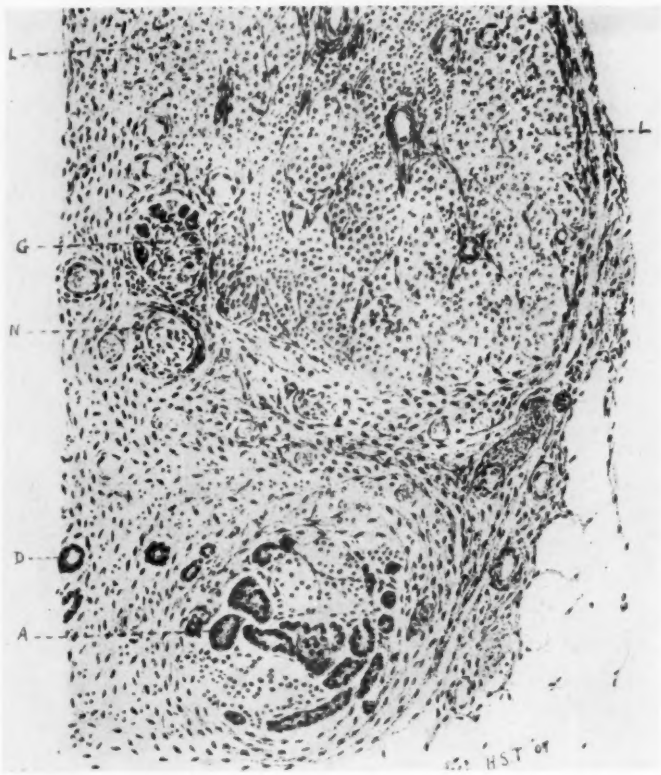

FIG. 2.—Section from pancreas of a dog containing a calculus. Courtesy Dr. Roy D. McClure. Portion of atrophied pancreas distal to obstruction of duct. *L*, degenerated parenchyma; *G*, ganglion with large nerve cells; *N*, nerve trunk; *D*, small branch of pancreatic duct; *A*, remnants of island of Langerhans.

glycosuria, when present, would suggest pancreatic disease, its absence in instances of pancreatic calculi is the rule.

Most of the operative cases have been between 35 and 50 years of age, though Thomason[69] reported a girl, age 14, and Sistrunk[65] a man, age 68. The history is usually of about 15 to 20 years' duration. Many authors have stated that pancreatic stones are much more common in men, but we find that 33 were men, 28 were women and in four the sex was not given. Only one of the 65 operative cases was a Negro. Stones have been reported in a Chinese woman.

Attacks seem to be brought on by fatty or oily foods. Schondube[61] reports the case of a man whose attacks were aggravated while he was sta-

tioned in Constantinople and eating Turkish food. Alcoholic excesses also seem to cause exacerbations.

Frothy, ill-smelling, grayish stools are rarely reported, but when present, suggest the correct diagnosis.

Diagnosis.—Prior to 1925, the preoperative diagnosis of pancreatic stones had been made in probably less than six cases. Since then the diagnosis has been made more frequently, though the percentage is still small. It is impossible from the case histories to state just how often it is made.

The main differential diagnosis is cholelithiasis. A few diagnostic criteria of pancreatic stones are listed briefly:

(1) Colic-like pain referred to the back or left epigastrium.
(2) Slight preponderance in males.
(3) Average age of patients around 40.
(4) Long intervals between attacks.
(5) Bilious vomiting and increased salivary flow.
(6) Midepigastric tenderness.
(7) Steatorrhea when present.
(8) Relative absence of jaundice; present in 27.6 per cent.
(9) Expulsion of calcium carborate or phosphate stones in the stools.
(10) Moderate loss of weight and strength.
(11) Glycosuria when present.
(12) Typical appearance of stones roentgenologically.

A positive diagnosis before operation can be made only roentgenologically. A duodenal catheter may be used in circumscribing stones in the head of the pancreas on the roentgenogram.

Treatment.—The treatment of pancreatic calculi is surgical removal in all cases except where there is diffuse calcification throughout the entire pancreas. The results reported following removal of stones are good, and several of the cases have been followed from five to 14 years without further attacks. There are relatively few postoperative complications.

For various reasons operation has not been performed in at least 28 cases diagnosed roentgenologically. Bernay[5] reports the case of a nun who had had three previous operations for ovarian and mesenteric cysts and adhesions, and radiographs now show stones in the head of the pancreas. Complete relief in an attack was obtained by the passage of a duodenal tube and she preferred this method of treatment to a fourth operation.

The use of hydrochloric acid for the elimination of calcium salts from the pancreas is urged by Loeper[42] and others. Loeper cites the case of a young diabetic with pancreatic stones, demonstrated roentgenologically, whose glycosuria fell from 180 Gm. per 24 hours to 36 Gm. after ten days of ten drops of hydrochloric acid before meals. Some of the stone shadows also disappeared.

Surgical Approaches.—There are several satisfactory approaches to the pancreas. Moynihan and Lisanti used a transduodenal incision for removal of stone in the duct of Wirsung, near the ampulla of Vater. The most com-

monly used approach, however, is through the gastrocolic omentum into the lesser peritoneal cavity. Link and others incised the mesocolon, and Link delivered the tail of the pancreas into the wound before opening the duct of Wirsung. Others have exposed the pancreas through the gastrohepatic omentum. The approach used depends, however, upon the location of the stones.

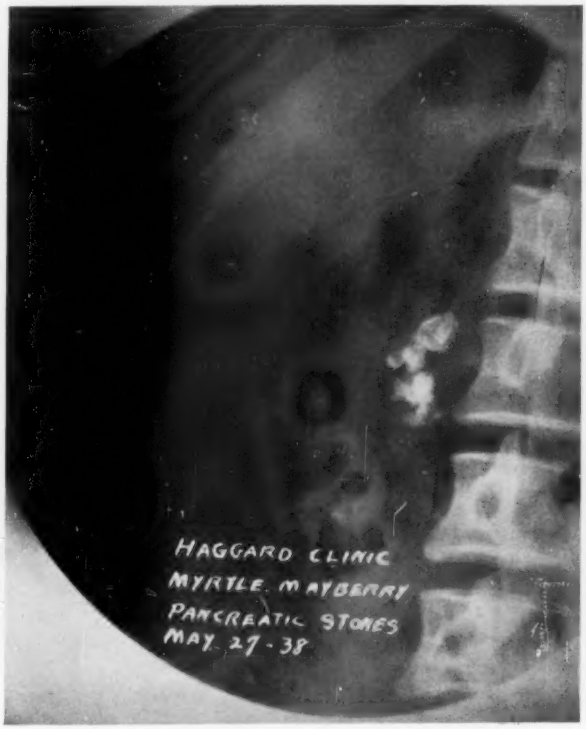

FIG. 3.—Author's case. Negative exploration elsewhere for gallstones. Roentgenogram afterwards showed pancreatic stones. Successful removal five weeks after exploration.

Pancreatic Fistulae.—Fistulae following the removal of pancreatic calculi usually close within a few weeks or months. Several are reported as having drained for many years, one as long as 14 years (Link), but all eventually closed spontaneously.

In our case drainage averaged about 300 cc. daily over a period of five months, with the exception of a week during which the fistula was apparently closed. There is usually little autodigestion of the skin.

Craft[19a] has shown experimentally that ephedrine definitely reduces the output of external pancreatic secretion. This is due to its vasoconstricting action and the fact that the pancreas is very sensitive to changes in the volume of blood flow (Babkin). A slight but definite decrease was noted in the case to be reported when ephedrine was given every four hours. It is possible that larger doses might have been more effective.

A diet rich in fats and proteins and low in carbohydrates was suggested by Wohlgemuth, in 1907, to aid in reducing pancreatic secretion. Pavlov proved that fats caused less secretion than proteins or carbohydrates. The administration of sodium bicarbonate was also advised. Clinical reports show an almost equal difference of opinion regarding the effectiveness of diet and alkalis in reducing the total output of the fistula. In our case there was no appreciable reduction during the period that the patient was taking this diet.

Case Report.—M. M., white, female, age 35, with a boy 15 and twin girls, age 5. Her history dated back 30 years (age 5) to repeated attacks of epigastric pain associated with vomiting and jaundice. The pain was severe enough to make catarrhal jaundice at that time unlikely. These attacks occurred every year or so until patient was age 11. There were no further attacks until the patient had her first pregnancy. She has had about 12 severe attacks, with jaundice with ten of them. Pain is knife-like, sudden in onset, and usually confined to the epigastrium. The pain required morphine and was not relieved by nitroglycerin.

FIG. 4.—Roentgenogram of the pancreatic stones after removal.

Two weeks after the onset of the present attack (April, 1938) cholecystostomy was performed at another hospital, although the diagnosis of pancreatic stones was made by palpation at the time of operation. Except for slight dilatation of the gallbladder and common duct, the biliary tract was normal. Lipiodol was injected through the drainage tube a week after operation, and the stones were seen to lie outside the biliary system, though close to the ampulla of Vater (Fig. 3). Drainage from the gallbladder ceased as soon as the tube was removed.

Examination, five weeks after this operation, revealed a healed wound with slight epigastric tenderness, but no masses. Blood sugar, glucose tolerance, uranalysis, and examination of the stool were normal.

Secondary operation was carried out five weeks after the first one (May 27, 1938). The gastrocolic ligament was incised and the duodenum mobilized and retracted superiorly. A vertical incision over the stones in the pancreas was then made and stones were removed in about a dozen pieces (Fig. 4). They were embedded in the substance of the head of the pancreas and lying in or around the duct of Wirsung. The stones seemed to be in a cavity and the duct was not definitely identified. The cavity was drained and except for the large amount of pancreatic drainage, the postoperative course was uneventful.

Several weeks after operation a catheter was placed in the fistulous tract and the drainage collected in a rubber container and measured. This allowed the mild excoriation of the skin to heal and was easily managed by the patient. The drainage varied from 300 to 600 cc. per 24 hours and was increased at night.

Following the local application of silver nitrate, the fistula closed for six consecutive

days, but was reopened. There was considerable pain in the left epigastrium and back at this time, which was relieved by reopening the fistula. About five months after operation the fistula was again cauterized with silver nitrate and has remained closed for the past two months, and there has been no pain, digestive disturbances, or glycosuria.

Roentgenologic examination, postoperative, showed several scattered, irregular areas of calcification at the level of the third lumbar vertebra. They were not felt in the pancreas after the mass of stones had been removed and should cause no further trouble.

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In the experimental laboratory, it has been known for some time that after pancreatectomy, the death commonly is due to conditions other than diabetes. Best and his collaborators have shown that the terminal stages of diabetes are due to hepatic insufficiency produced by fatty infiltration of the liver. Several cases have been presented in this country by Snell, Comfort and Rosenberg, in which the presence of fatty infiltration of the liver has been proved microscopically. Lester Dragstedt was the first to show that a pancreatic extract of a hormonal nature controls this deposition of fat in animals. This preparation has been called "lipocaic" and is an extract obtained from the pancreas of cattle. Clinical confirmation of the action of this pancreatic extract is found in the interesting case presented by Rosenberg before a meeting of the American Gastro-Enterological Association in May, 1938. He was able to show histologically that fat disappears from hepatic cells when "lipocaic" is administered.

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It is logical to assume, therefore, that calculi in the pancreatic duct will produce variable degrees of pancreatic atrophy, depending on the degree of obstruction of the duct. Support for this contention was presented by Dr. Joseph Mayo in a symposium on pancreatic disease in 1936. He called attention to the fact that of nine cases in which pancreatic lithiasis was found

at postmortem examination at the Mayo Clinic, in three, atrophy of the pancreas was of sufficient degree to produce changes in the liver. It seems apparent, therefore, that there is a necessity for removing pancreatic calculi. That in some cases it will be possible to do this with comparative ease is evidenced by a case which I presented at the same symposium, demonstrating that some 15 or 20 pancreatic calculi, varying in diameter from 5 to 8 Mm. had been removed from the substance of the pancreas by cutting directly down on them and, in the subsequent course of the disease, it was not apparent that incisions had been made in the pancreas.

These are, however, cases in which calcification may be scattered throughout the entire pancreas. These calcareous accumulations are probably so numerous and so inaccessible that they could not possibly be removed. They usually develop in the course of healing of fatty necrosis and are not, therefore, true pancreatic calculi which could obstruct the duct.

One should not hesitate to expose the pancreas for surgical exploration. I have seen cases in which an operation had been performed previously on the pancreas for a suspected adenoma, in which it was necessary to explore it again; it is surprising that such a procedure is not more difficult than it is. Rynearson and I have reported recently a case in which I performed an almost complete pancreatectomy; this patient previously had been operated on twice for pancreatic disease.

MESENTERIC LYMPHADENITIS

WALTER D. WISE, M.D.
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AN ACCURATE history of surgery should contain a chapter on changing pathologic conditions found in the abdomen. The increase in the number of instances of appendicitis, the greater frequency of biliary tract disease in the Negro, the increased incidence of diverticulitis of the sigmoid and other parts of the colon, the apparent increase of abdominal malignancies, and the comparatively recent observations on ileitis are subjects in point and there are perhaps many others. Whether there are greater numbers of these conditions, or the more faulty observations of the past make it appear so, it is not easy to say. It is difficult to believe that such a condition as ileitis, as it is described to-day, could have been overlooked at operation or necropsy until so recently.

A somewhat similar situation obtains in the instance of mesenteric lymphadenitis, a condition that has, it is true, been known since Sydenham's time or earlier, but only recently has it been recognized that there is a nontuberculous variety occurring with considerable frequency. Some surgeons have seen a great number of cases, yet it is practically unknown to others familiar with abdominal surgery and with large practices.

The recent appearance of numerous comprehensive articles on mesenteric lymphadenitis makes a lengthy consideration of this subject inappropriate, and I refer those interested to a short bibliography which will, in turn, give a very complete list of references. I should like, however, to direct attention to this condition, to add nine personal cases to those in the literature, and to call particular attention to certain manifestations in my small series, the most striking of which included a seasonal incidence and uniform recovery after appendicectomy.

Etiology.—Prior to 1937, my experience with mesenteric lymphadenitis of unexplained origin had been limited to a few instances, scattered over a long period of time, occurring mostly in children. They were thought to be tuberculous and the subsequent care of the patients was based upon this hypothesis, though one node which was removed was pronounced nontuberculous.

In the late summer of 1937, a striking experience directed attention and interest to the subject under discussion. Four acute or subacute cases were seen between August 15th and September 14th, with two of the more chronic type during the same season; one on August 1st and one on September 10th. In spite of a sharp lookout, no others were then seen until August 27, 1938, a second one being seen September 1, 1938. An exception is Case 9, B. B., age 15, who was seen May 24, 1938, but was not typical of the others described in this report because he suffered with a chronic *Streptococcus viridans* throat infection, and, all his life, had been allergic to most of the common

TABLE

ANALYSES OF NINE CASES OF

Patient Date	History of Respiratory Tract	Onset	Physical Signs	On Admission Temp.	Pulse	Resp.	Leukocytes Percentage Polys.
H. B., male, age 43 yrs. Aug. 1, 1937	No recent respiratory infection. T and A out.	Pain both lower quadrants, esp. left; occas. passing to scrotum; no nausea; no vomiting. Moderate frequency; nocturia and urgency. Mild chill	Tenderness in both lower quadrants; no rigidity	99.2—	88—	20	13,350—85% 9,150—75% 10,600—71% 6,450—64% Serology, neg.
P. S., male, age 15 yrs. Aug. 18, 1937		Pain began in epigastrium, passed to R. L. Q., 24 hrs. duration; no nausea; no vomiting. Pain on urination	Tenderness over McBurney's Pt. Rebound tenderness; tenderness over right lumbar region. Throat—neg. Lungs—neg.	101	—100—	26	6,100—69% 5,400—57% 7,200—74% Serology, neg.
O. LeM., male, age 9 yrs. Aug. 25, 1937	Previous colds and sinus trouble. No sore throat. T and A not removed	Pain one day. Vomiting	Tenderness lower half of abdomen. Slight rigidity	99	— 86—	20	20,000—92% 14,100—93%
H. H., male, age 7 yrs. Sept. 2, 1937	No recent colds or sore throats, though many earlier. T and A out in 1936	Recurrent pains in lower right side for 3 days. Slight nausea; no vomiting. 1½ yrs. ago similar attack	Moderate tenderness over McBurney's Pt. Moderate distention. No rigidity. Throat—neg. Lungs—neg.	98.4—	98—	26	7,700—54% Lymphocytes —43% Serology, neg.
F. Z., male, age 25 yrs. Sept. 10, 1937	Recent head colds and sinus infections. T and A out 12 yrs. previously	Recurrent pains in L. L. Q. and R. L. Q. for 6 days. No nausea; no vomiting. Frequency in urination, nocturia, urgency	Tenderness in L. L. Q. and R. L. Q. Mostly to right side of umbilicus. Slight rigidity	97.3—	76—	20	5,800—74% 6,500—73% 8,500—63% 5,800—68% Serology, neg.
M. D., female age 8 yrs. Sept. 14, 1937	No recent colds. Quite a number some time before operation. T and A out	Pain on right side constantly for 4 days. Moderate nausea; no vomiting	Moderate tenderness over McBurney's Pt. No rigidity. Throat—neg. Lungs—neg.	101	—104—	24	10,700—66% Serology, neg.
M. L., male, age 10 yrs. Aug. 27, 1938	Frequent colds and sore throats	Pain in R.L.Q. and central abdomen. No nausea; no vomiting. No urologic symptoms. Similar attack 1 yr. ago	Pain on deep pressure, R.L.Q. No rigidity	100	—124—	22	9,750—79% Serology, neg.
J. C., male, age 6 yrs. Sept. 1, 1938	No history of colds or throat infections. T and A not removed	Constant pain in R.L.Q., 14 hrs. duration; also pain in umbilical region. Nausea and vomiting	Tenderness on deep pressure over McBurney's Pt. No rigidity	102.3— 104	120—	22	6,250—94% Serology, neg.
B. B., male, age 15 yrs. May 24, 1938	Chronic *Streptococcus viridans* infection of throat. T and A out	Ten days ago acute throat infection. Temp. 104°; 7 days ago, pain in R.L.Q. began; temp. 101°; 2 days' diarrhea. Patient suffers almost constantly with *Streptococcus viridans* throat; asthma; allergic to nearly all foods, dust, *etc.*	Moderate tenderness over McBurney's Pt. Slight rigidity	99 101	—100—	22	11,700 11,600 7,300 Serology, neg. Serology, neg.

I

MESENTERIC LYMPHADENITIS

Urine	Roentgenologic Examination	Operation	Pathologic Report	Follow-Up Report
Albumin—trace. Occas. W.B.C.	Calcified nodes. Pyelogram normal	No operation		October, 1938: Health fairly good; occasional abdominal pain
Occas. W.B.C. and R.B.C.	Calcified lumbar no les. Pyelogram negative	Right paramedian incision below umbilicus. Appendix not definitely inflamed. Calcified node about 2 cm. in diameter in mesentery, surrounded by smaller inflamed nodes. Calcified node and several smaller nodes removed	Appendix—edematous. Mucosa scarred. Leukocytic infiltration. Subacute appendicitis. Lymph nodes: (1) Round cell infiltration. No tubercles. (2) Large node calcified	No answer to questionnaire
Negative		McBurney incision—Appendix perhaps slightly inflamed but not looking to be responsible for symptoms. Mesenteric lymph nodes enlarged, soft, pink, averaging about 1 cm. in diameter, some larger, some smaller. Small amount of clear free fluid in abdomen. No nodes removed	Chronic catarrhal appendicitis	October, 1938: Health excellent. Colds but no abdominal symptoms
Occas. W.B.C.		McBurney incision—Appendix rather innocent looking grossly. Lymph nodes and fluid similar to Case 3. No nodes removed	Appendix hyperemic, edematous, mucosa thick. Follicular lymphoid hyperplasia. Moderate leukocytic infiltration. Acute appendicitis	October, 1938: Had one mild attack after operation. Health now excellent, but gets head colds
Albumin: trace. Occas. W.B.C.	Calcified nodes. Pyelogram negative	McBurney incision—Appendix innocent looking. Some lymph nodes about as in Case 3. Others calcified, but only about 1 cm. in diameter. Four to 6 inches of terminal ileum bound tightly to posterior abdominal wall. No nodes removed	Submucosa—marked fibrosis. Mucosa infiltrated with small mononuclears and eosinophils. Chronic appendicitis	October, 1938: Health excellent. No further attacks. Has gained 20 lbs.
Negative		McBurney incision. Findings similar to those in Case 3. No nodes removed	Appendix edematous. Markedly infiltrated with polys., eosinophils. Acute appendicitis. Culture of fluid negative	October, 1938: No further trouble. Has had colds but no abdominal symptoms
Negative		McBurney incision. Findings similar to those in Case 3. No nodes removed	Follicular lymphoid hyperplasia throughout mucosa. Leukocytic infiltration is confined to mucosa and most of cells are mononuclears. *Diag.:* Early acute appendicitis. Abdominal fluid culture sterile	November, 1938: No further trouble. Health excellent
Occas. W.B.C.		McBurney incision. Findings similar to those in Case 3, with much free clear fluid. No nodes removed	Follicular lymphoid hyperplasia. Germinal follicles hypertrophied. Infiltration cells are mononuclears. *Diag.:* Marked lymphoid hyperplasia and chronic inflammation of the appendix. Abdominal fluid sterile	November, 1938: No further trouble
Occas. W.B.C.		Spinal anesthesia. McBurney incision. Appendix was tied down, not acutely involved. Large, liver-colored nodes in meso-appendix. None in mesentery of ileum. Appendix and large node removed	Appendix short, firm, cord-like. Mucosa and lumen entirely obliterated by edematous scar tissue. There is infiltration with leukocytes—small mononuclears and eosinophils. Obliterated appendix with chronic inflammation. Portion designated for pathologic laboratory was lost. Node cultures negative in 2 laboratories, for bacteria and viruses	General health better. Still has throat infection; allergic reactions; no further attacks of abdominal trouble

foods, dusts and pollens, *etc.*, and who had had abdominal symptoms at various times.

The records at the Mercy Hospital, Baltimore, show, for 1937 and to November 20, 1938, five other patients diagnosed as having this condition, which, however, were not proven by operation, all occurring in the late summer and fall except one which was seen in April, 1937. A sixth case, proven by operation, occurred in August, 1938. All of this group were in children.

Brennemann, in 1921 and again in 1927, directed attention to the abdominal pains of children suffering with throat infections, and many writers since then have attempted to show association of the enlarged mesenteric nodes and upper respiratory infections. Six of my nine patients gave histories of such infections; in one there is no information on this subject, two had no such history. However, quite a number have been reported as occurring in patients without any evidence of throat or upper respiratory tract involvement or history of recent trouble of this nature. As this condition occurs mostly in children, it is not surprising to find a history of upper respiratory infection no matter what the "present complaint."

Appendicitis as a cause, or as an associated condition, is discussed at perhaps greater length than any other factor.[1, 2] The beneficial effects of removal of the appendix in a large percentage of cases have been described by Royster quoted by Freeman,[3] Brown,[4] Strombeck,[5] and many others.

Other suggested causes of nonspecific adenopathy include (mention and discussion of) numerous conditions local and general. The list embraces in addition to infections of the respiratory tract, appendicitis and tuberculosis, all of the granulomata, typhoid fever, undulant fever, nonspecific inflammations of the intestines, intestinal influenza, intestinal stasis with absorption of histamine or other products and intestinal parasites, malignancies, Hodgkin's disease, leukemias, allergies, vitamin deficiencies, food poisonings, and metal or vegetable poisons. It is seen from this confusion that it is not clear whether the enlarged nodes are a result of an infection, a toxin, a virus or organic or inorganic poisons, but specific tests, and the lapse of a little time, cause many of the suggested factors to be discarded. The evidence seems to point to a virus or a toxin but has not been definitely proven, and infection in some cases has been proven present. White and Collins,[6] who have made careful studies of the bacteriology, feel that the condition is a virus disease and offer the somewhat startling but interesting suggestion that it may represent an abortive type or stage of infantile paralysis. Their grounds for advancing this possibility are their belief that it is a virus disease, a similarity of lymphadenopathy found in cases of poliomyelitis, with some similarity of the abdominal symptoms. Whether the recent work of Trask and Vignec,[7] demonstrating the virus of poliomyelitis in the human stool, will further this suggestion must await more study. These authors kindly offered to examine a stool from one of my more recent cases of adenitis, but, unfortunately, did not at the time have monkeys available for the work.

As suggested earlier, it is well established that most of these cases occur

in children but occasionally one sees them in the adolescent and the adult. The ages of the patient with acute symptoms in my series were 15, 9, 7, 8, 10, 6, and 15. Cases designated chronic were in adults, one age 25, the other 43. There was only one female in the series.

Pathology.—While most surgeons have been wary of removing nodes, even for biopsy, for fear of the presence of Streptococci and fear, in the exaggerated cases, of interference with intestinal blood supply, yet quite a number have been studied. These have failed to throw much light on the subject, either bacteriologically or pathologically. All except an occasional specimen have been sterile, after all types of culture, and pathologically they showed only an hyperplasia and sometimes hemorrhagic infiltration, suppuration and calcification. Single and conglomerate masses of nodes of great size have been described. A few instances of streptococcic invasion has been reported but these are rare exceptions and not the rule. My patient with severe chronic throat infection showed sterile nodes. Some authorities still feel that the so-called simple adenopathy may be a phase of tuberculosis.

Symptomatology.—The symptoms in the early cases are in most instances mild. Advanced stages of the condition may have nodes or groups of nodes visible or palpable, some of almost unbelievable dimensions, with the expected accompanying manifestations. Suppuration and rupture lead to a localized abscess or general peritonitis. Except for one patient, who had a high temperature and who appeared quite ill, the symptoms in the group herewith reported were more subacute than acute, some possibly being chronic with exacerbations. Those classed as chronic had symptoms similar to the mild, acute ones and were so classified because of the demonstration of calcified nodes. The complaints were of abdominal discomfort, usually more pronounced in the right lower quadrant, occasionally similar symptoms on the left side, moderate tenderness, little or no rigidity of overlying muscles, varying degrees of nausea and vomiting, varying degrees of temperature and leukocytosis—the counts ranging from 5,000 to 20,000, with polymorphonuclear counts from 54 to 95 per cent, most of them between 60 and 80 per cent. In most of the patients the symptoms and signs were quite suggestive of a low grade, somewhat atypical, appendicitis, and the first few were operated upon for this condition. Towards the last of the series, the correct diagnosis was tentatively made, but, fearing a mistake, it was thought safer to operate. In no patient, except the one with chronic throat infection, was there clinical evidence that upper respiratory tract disease existed at the time of abdominal symptoms, but most of them had suffered in the recent past with colds or sinus infections. While there was no time in most instances for complete studies, four of the patients were referred by a careful pediatrician, all were seen by one or more consultants, including appropriate specialists, as urologists and others. Three had mild urinary symptoms.

Operative Pathology.—In no case of the series was the appendix plainly guilty on gross inspection, though all showed microscopic changes—two being called acute. The outstanding operative findings were enlarged lymph nodes

in the mesentery of the small intestine, in some instances widespread; in others confined to that portion of it leading to the terminal ileum. In some the nodes of the meso-appendix were involved. In three cases, two classed as chronic, and one with more acute symptoms, there was calcification of nodes. In two of these, operated upon, there were also enlarged noncalcified nodes seemingly involved in a fresh inflammatory process. The noncalcified nodes, when seen at operation, varied in size from one-half to one and one-half centimeters in diameter. They were pinkish-gray in color and soft. The calcified nodes were much larger. The association of the two conditions would seem to indicate stages of the same process and call for the addition of the calcified group to Rosenberg's[8] clinical classification of: (1) Chronic; (2) acute; (3) acute fulminating; and (4) acute suppurative mesenteric lymphadenitis. Wilensky and Hahn[9] called attention to this class in 1926.

In several instances there was a considerable amount of clear free fluid in the abdomen, the largest amount being in the patient with the highest temperature. This fluid was uniformly sterile, as has been noted by others. There were no suppurative nodes in this series nor were there any excessively large ones or masses. Noncalcified nodes were removed in two instances. In one, the specimen was lost in transit to the pathologic laboratory but it was examined bacteriologically. The report of the pathologist in the other was "round cell infiltration—negative for tuberculosis." A node from one of these patients was cultured by our laboratory with negative results bacteriologically, and The Johns Hopkins University School of Hygiene pronounced it negative for filtrable viruses.

Diagnosis.—A history of any type of upper respiratory infection accompanied by, or followed by abdominal symptoms of an atypical appendicitis may suggest the diagnosis, but obviously too much dependence upon this would lead to serious error. Laboratory examinations have been of little aid. Klein[10] suggests that turning the patient from side to side causes a shift of the area of tenderness, differing in this from appendicitis, as the cecum is more fixed than the mesentery. Ochsner and Murray[11] stress the presence of tenderness along the root of the mesentery, i.e., on a line extending from McBurney's point upward and to the left of the umbilicus. Mead[12] believes the acute form cannot be differentiated from acute appendicitis.

A recent case, a boy, age 13, with a history of recent colds, had more or less general abdominal pain, and tenderness in the right iliac fossa, midzone and to the left, with temperature of 99.5° F., leukocytes 6,000. A tentative diagnosis of mesenteric lymphadenitis was made but he was found to have a very definite appendicitis, with no adenitis.

Treatment.—There is no specific medical treatment and for the extensive node involvement surgical removal is, of course, out of the question. Calcified nodes may be removed, if removal can be accomplished without wounding mesenteric vessels and if one does not fear peritoneal contamination. This, like the removal of noncalcified nodes, is usually for biopsy rather than treatment. The appendix has been removed: (1) Because of mistaken diagnosis;

(2) because there is a belief, by some authorities, that the appendix is the cause of the condition; and (3) because by experience, as in this series, it has been productive of good results. Simply opening and closing the abdomen has been advocated, as in cases of abdominal tuberculosis, and it is claimed that the results have been beneficial. Naturally, evident foci of infection should be remedied, a proper dietary regimen carried out, and all other appropriate hygienic and medical measures seen to. No attempt has been made in this paper to consider suppurative cases with abscess formation or general peritonitis, which, of course, present a different picture and need treatment that includes drainage.

DISCUSSION

No conclusions can be reached from the findings in this small series, but attention may be called to a few debated questions:

1. Of the eight cases questioned, six had histories of recent or fairly recent upper respiratory infections, two had not, in one there is no record. As most of the patients were children such a history may be a coincidence rather than cause and effect. Four patients (Cases 3, 4, 6 and 9) have had subsequent upper respiratory infections without abdominal symptoms.

2. Two (Cases 4 and 7) had histories of symptoms recurring for a year or more (as did a case seen five years ago, not included in this series). This would seem to cast doubt on the poliomyelitis hypothesis, as that disease does its damage promptly, does not recur and confers immunity. None of this series developed any paralyses nor have I seen mention of any others who did.

3. One patient suffered unusually severe allergies and had a chronic *Streptococcus viridans* throat infection. Since appendicectomy, he has had no more abdominal symptoms, yet microscopic examination showed the appendix to have an obliterated lumen. A markedly enlarged, liver-colored lymph node removed from the meso-appendix was sterile, and no filtrable viruses could be demonstrated.

4. Though this seasonal incidence has not been corroborated by all other observers, in my series it was paralleled by other admissions to Mercy Hospital with one exception, one of the unproven cases. This brings up the question of food, atmospheric or other seasonal factors.

5. In this series, temperatures and blood counts varied too greatly to be characteristic.

6. Eight patients had appendicectomies performed, seven had been entirely relieved. One patient cannot be traced. The one who was not operated upon continues to have abdominal symptoms. One patient, Case 9, remains ill of the *Streptococcus viridans* throat infection and allergic manifestations but has no more abdominal symptoms. Eight of the nine patients were private and, barring the unforeseen, can be followed indefinitely.

It is interesting to speculate why the removal of rather innocent looking appendices should produce such a large percentage of symptomatic cures. Strombeck,[5] who studied 40 patients, averaging nearly four years after opera-

tion, found 87 per cent had symptomatic recoveries. My small series of nine, with eight operated upon and seven accounted for, shows symptomatic recoveries in all who had the appendix removed. Two have been followed for a few months and five for over a year. These results are not reported by all writers and do not allow of a definite conclusion.

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DISCUSSION.—DR. I. M. GAGE, New Orleans: For several years we have been interested in mesenteric lymphadenitis because we see a very high percentage (about 60 per cent) of enlarged mesenteric lymph nodes in our cases of chronic appendicitis.

We have divided our cases into two main groups: (1) Those cases in which symptoms are acute (simulating acute appendicitis), and are designated as acute mesenteric lymphadenitis; and (2) those in which symptoms are subacute or chronic (simulating chronic appendicitis), and are designated as chronic mesenteric lymphadenitis.

The first group, in the majority of instances, is diagnosed as acute appendicitis. If a patient presents a clinical picture of acute appendicitis and at operation the appendix appears normal, one should always investigate the mesentery of the terminal ileum for mesenteric lymphadenopathy. In the acute form of mesenteric lymphadenitis, the lymph nodes are edematous and somewhat pinkish in color, with an immediate edema surrounding the lymph nodes. This group is easily recognized and the only discomforting factor is that the etiology is obscure.

The second group, is the type that we have been interested in for over 12 years. Chronic mesenteric lymphadenitis occurs in about 60 per cent of our cases of chronic appendicitis. The patient presents a clinical picture of chronic appendicitis and at operation we find evidence of disease in the appendix and also a concomitant enlargement of the mesenteric lymph nodes. The nodes increase in size as the base of the mesentery is approached. In all

operations for appendicitis both chronic and "acute nonperforating," the terminal ileum and its mesentery is routinely inspected for a distance of 24 to 36 inches. This probably explains the high incidence of chronic mesenteric lymphadenitis found in our cases. Incidentally, we have also encountered a Meckel's diverticulum in over two per cent of our patients.

We investigated 65 cases of chronic lymphadenitis in an attempt to determine the age incidence, etiology, pathology, symptomatology, and diagnosis. The age incidence was as follows: Forty-five per cent occurred between the ages of 11 and 15; 34 per cent between the ages of 21 and 34; and 0.02 per cent between the ages of 4 and 10. The largest group, therefore, occurred in the adolescent period, i.e., 11 to 15 years. The ratio of males to females, was almost 4 to 1.

The bacterial flora of the enlarged nodes was investigated by culturing the nodes, then identifying the type of organism found on culture. The lymph nodes were removed by the most careful aseptic technic. Half of the gland was then cultured and the other half was fixed and stained for histologic and bacteriologic study. The node was ground up in a mortar, in sterile sand, and then cultured in brain broth medium. In 67 per cent of the node cultures, the enterococcus was obtained in pure culture. In 27 per cent a gram-positive Diplococcus in association with the colon bacillus was found and in one case, a gram-positive Diplococcus in association with a Streptococcus was found. The enterococcus therefore was found in 93 per cent of the nodes cultured. Due to negative agglutination tests, and negative sensitivity tests to intradermal injection of the vaccine made from the enterococcus, we, therefore, were not able to draw definite conclusions, or could we assume that the enterococcus was the etiologic factor. However, the high incidence of the enterococcus (93 per cent) in cultures was suggestive, that the enterococcus may play a part in the etiology of this clinical condition.

Infections of the node were stained with hematoxylin and eosin, Mallory's connective tissue stain, and Goodpasture and Lilly's modification of the gram stain for bacteria. The majority of the nodes revealed rather definite changes in their architecture, characterized by fibrosis, and hyperplasia of the recticulum cells. The lymph follicles were decreased in size showing marked atrophy in some instances. The fibrosis occurred mainly in the capsule and trabeculi, however, thin strands were found penetrating the parenchyma. Bacteria (Cocci) were demonstrated in all of the nodes studied, varying in number from a few to large groups.

Pain occurred in 100 per cent of our cases. It was cramp-like or colicky in 60 per cent, dull or aching in 23.8 per cent, and knife-like in 16 per cent. Nausea occurred in 41.5 per cent, vomiting in 20 per cent, and constipation in 10.7 per cent. Tenderness was present in 100 per cent. It was demonstrated along the root of the mesentery of the small bowel, i.e., obliquely across the abdomen from the right iliac fossa, across the umbilicus towards the left costal angle, in 72.4 per cent. Pain was localized at McBurney's point in 23 per cent and at Land's point in 32 per cent. Rigidity of the abdominal muscles, especially in lower right quadrant, occurred in 23 per cent.

The diagnosis of chronic mesenteric lymphadenitis is made on the above clinical signs and symptoms. However, chronic appendicitis cannot be eliminated. Therefore, in the majority of our cases the diagnosis of chronic appendicitis with concomitant chronic mesenteric lymphadenitis was made. If a patient presents the clinical manifestations and physical findings of chronic appendicitis and in addition pain on pressure along the route of the mesentery of the small bowel is present, a diagnosis of chronic mesenteric lymphadenitis is also made. In all of our cases an exploration is made, the appendix re-

moved, and the mesenteric lymph nodes investigated. Therefore, we are convinced that all cases should be operated upon and the appendix removed.

A questionnaire was sent out to the 65 cases that were studied. A reply was received from only 22 patients, and of these only 60 per cent stated that they were completely relieved of their symptoms. I saw some of these patients later and, even though they stated that they were not relieved of their symptom, on examination it was found that they only had a sensitive scar. The other clinical manifestations complained of preoperatively had entirely cleared up.

Dr. Walter D. Wise, Baltimore, Md. (closing): One point that has been brought up in this discussion, I think, is very interesting, that is the high percentage of cases of lymphadenitis seen by Dr. Gage, because in most reports it is stated that with appendicitis the mesenteric lymph nodes are not much involved. We do see a certain amount of enlargement in an occasional case, but that is not the type I am talking about and it may be that there is something in the South, in Dr. Gage's district, that produces this condition. It is interesting to know that he has found some constant bacteriology in these nodes.

As regards diagnosis, I recently had a case that showed the symptoms of what looked to be a low-grade mesenteric lymphadenitis with tenderness along the line running from McBurney's point upward and to the left of the umbilicus and made the diagnosis and operated, and he had ordinary acute appendicitis and no lymphadenitis. It is interesting to note that practically all Dr. Gage's cases were relieved by appendicectomy. In one case of mine the appendix was fibrotic, clearly not active, yet the patient was entirely relieved.

THE RELATION OF CHRONIC INFLAMMATION AND ESPECIALLY LYMPHOGRANULOMA INGUINALE TO THE DEVELOPMENT OF SQUAMOUS CELL CARCINOMA OF THE RECTUM

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THE apparent importance of chronic inflammation as an etiologic factor in the development of squamous cell carcinoma is well known. What the exact causative factor in chronic inflammation consists of is not so clear. Chemical inflammation as in aniline dye workers, betel nut chewers and tar workers; specific inflammation as in luetic ulcers or lupus; bacterial inflammation as found in chronic bone sinuses; simple mechanical inflammation as the effect of jagged teeth on the cheek or gallstones in the gallbladder; and the scar tissue changes resulting from burns, roentgen ray or radium, present only a partial list of varying inflammatory factors favoring the development of squamous cell carcinoma.

Squamous cell carcinoma of the anus is rather uncommon and comprises less than 2 per cent of the cancers of the rectum. In general its development in relationship to chronic inflammation has not been stressed in the literature. A long standing dermatitis ani has been present in a few cases, but no particular etiologic emphasis has been placed on this rather common condition.

The relationship of fistula-in-ano to the development of squamous cell carcinoma has received some attention in the literature. In 1934, Mummery said that it was very doubtful whether conditions such as chronic fissures in the rectum or openings of chronic fistulae are an important factor in the origin of cancer. He reports two fistulae of 30 years' standing without cancer formation. Daniel Jones says it is useless to state that cancer starts in hemorrhoids, fistulae, ulceration or strictures of the rectum for there are so few cases where it can be proved. Buie and Burst, however, reported 36 cases of squamous cell cancer of the anus of whom four had a history of a fistula of six months' or more standing. Curtice Rosser reported from the literature 21 cases of cancer of the rectum associated with chronic fistulae. He believes that in eight of the patients the fistula acted as an etiologic agent.

In a series of over 500 cases of rectal fistula, we have seen but one instance where squamous cell carcinoma apparently, unquestionably, developed in the chronic inflammatory tissue of the fistula.

Case 1.—The patient was a white male, age 35, who came to us in the fall of 1926, with the history of having had a fistula-in-ano for several years. He showed a horseshoe fistula with several openings, the appearance of the lesion being that of tuberculosis,

although repeated biopsies and guinea-pig inoculations failed to prove it. The sections showed chronic inflammatory granulation tissue without giant ecIls but with a marked scar tissue reaction (Fig. 1). The patient entered the hospital several times during the following three years for drainage of abscesses. An internal opening of the fistula was never found. In October, 1929, he had a new abscess develop and after drainage of it he went to Arizona for a few months. While there a mass developed in the left ischiorectal fossa and he returned to Chicago in March, 1930. In April, 1930, he entered the hospital and a biopsy from the indurated area surrounding the fistula showed squamous cell cancer (Fig. 2). This was treated with radium without result. In July, 1930, a colostomy was performed and a month later a suprapubic cystotomy was necessary, due to involvement of the urethra. He died April 21, 1931. The autopsy showed a squamous cell carcinoma of the perineum and left thigh with destruction of the anal margins, carcinomatous infiltration of the small pelvis, and carcinomatous metastases to the left lung.

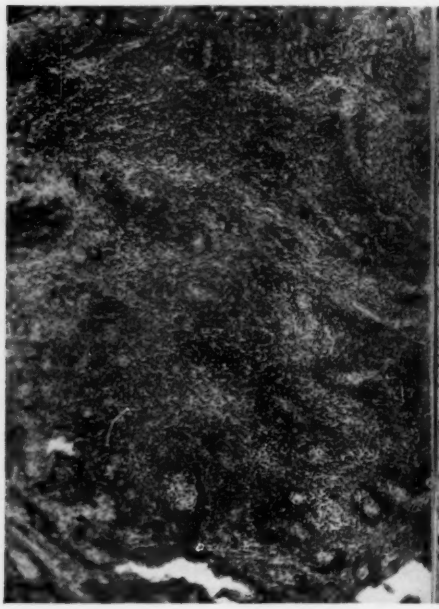

FIG. 1.—Case 1. Biopsy of chronic inflammatory tissue.

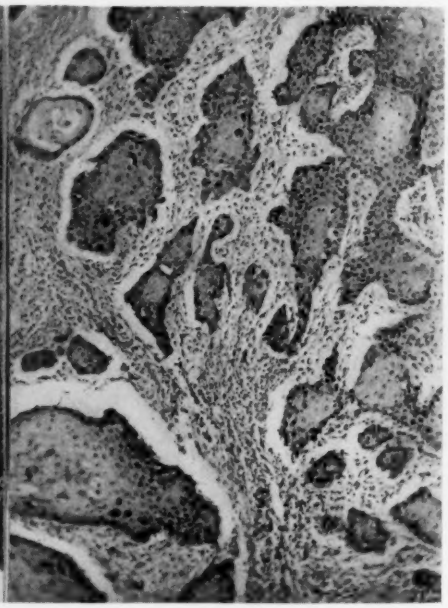

FIG. 2.—Case 1. Biopsy of tumor developing in chronic fistula.

In spite of this significant case, it seems apparent that carcinoma of the squamous cell type very infrequently takes its origin in fistula-in-ano.

Let us now consider the relationship of lymphogranuloma inguinale involving the rectum to the development of carcinoma. For many years those of us who have been working in large charity hospitals have seen many rectal strictures beginning as submucous infiltration and superficial ulceration and finally developing into fibrous strictures of the rectum. They continued to discharge blood and pus and to be frequently complicated by perirectal abscesses with continuing fistulous tracts. These lesions, due to the work of Frei, Nicolas, Favre and others, were finally classified as rectal manifestations of lymphogranuloma inguinale. We have had the opportunity of examining over 300 such lesions.

The first patient who developed a carcinoma in the fistulae draining from this type of rectal lesion was seen 20 years ago, before a specific diagnosis of lymphogranuloma inguinale could be made by the now established Frei test.

Case 2.—The patient was a female, white, age 45, who had been under our care for several years with a typical, and relentlessly developing, stricture of the rectum with all the well known characteristics of the lesion. Colostomy was advised but refused. Perirectal abscesses and draining fistulae into the ischiorectal fossa developed. She was miserable and nothing we did for her seemed to help in any way. After several years of suffering, induration of a progressive nature followed by a sloughing crater developed in the ischiorectal fossa at the site of the fistulous tracts. This lesion was squamous cell carcinoma.

During 1938, we have lost two patients from squamous cell carcinoma, each of whom had advanced lesions of lymphogranuloma inguinale in the rectum. Due to the fact that we had both of these patients under our care for several years and an autopsy was obtained in both instances, we have felt it worthwhile to call attention to the possibility of cancer formation in prolonged lesions of lymphogranuloma inguinale.

Case 3.—E. H., white, married, age 29, came to us in 1925, at which time she was pregnant for the first time, and was complaining of bleeding from the rectum. Examination at this time showed very slight induration of the mucosa and superficial ulceration on proctoscopic examination. A smear showed no gram-negative Diplococci, and cultures from the ulcerated area revealed no significant findings. The lesion was recognized as a venereal one and of the type we now know to be lymphogranuloma inguinale. A Wassermann was made, which was negative. There was no inguinal adenopathy of moment or lesions in the vagina. She was given weak silver nitrate enemata and a course of antiluetic treatment, although we felt sure both clinically and etiologically that the lesion in the rectum was not luetic.

As time went on, the lesion in the rectum became more pronounced; definite narrowing of the rectum was present, and on vaginal examination, the rectum as high as the posterior fornix could be felt as a fibrous cord. Pus and blood were discharged from the rectum. On September 29, 1928, four years later, she entered the Presbyterian Hospital, complaining of ribbon-like stools and discharging sinuses around the rectum. At this time, the general physical examination was negative except for a marked anemia. The Wassermann was negative. A rectal examination showed numerous hard nodules resting upon the leather-like base of the submucosa. The rectum was movable on the surrounding parts. The lumen of the rectum was markedly constricted. There were two complete rectal fistulae. Biopsy from the area of the rectal lesion showed round cell infiltration.

The patient did not return to the hospital for nine years, although during that period she had been given many intravenous injections of potassium and antimony tartrate with little effect.

In 1935, ten years after the onset, the patient developed recurrent diarrhea and abdominal cramps. She had lost weight and complained of pain over the sacrum. Her heart was enlarged and there had developed a loud systolic and diastolic murmur. She was tender over the descending colon and lower abdomen. The Frei test with several different antigens was positive. The skin around the anus was lacerated. Fistulous tracts were present. There was a large abscess in the left ischiorectal fossa running forward and involving the left labia. This was opened and drained with some relief of pain.

Following the drainage of this large abscess, the patient consented, for the first time,

to have a colostomy performed. This was carried out and she left the hospital on the sixteenth day. Following diversion of the fecal stream she was much more comfortable, gained 50 pounds in weight during the next few months, and was able to return to work.

She remained free of symptoms for two years but, in July, 1937, she returned to the hospital complaining of severe pain in the rectum and perineum, and loss of weight. She was anemic. The colostomy was functioning. On examination, a firm, hard mass was present in the perineum and rectovaginal septum. This was incised, pus evacuated, and tissue taken for biopsy, which was diagnosed squamous cell carcinoma. Profuse drainage continued. She became progressively weaker, and developed edema of the ankles. The inguinal nodes became palpable and hard. On December 3, 1937, she died. *Diagnosis:* Squamous cell carcinoma developing on lymphogranuloma inguinale of the rectum.

FIG. 3.—Case 3. Gross specimen of squamous cell carcinoma developing on lymphogranuloma inguinale.

FIG. 4.—Case 3. Biopsy of tumor.

Autopsy.—Dr. Carl Apfelbach. *Anatomic Diagnosis:* "Squamous cell carcinoma of the rectum (Figs. 3 and 4); carcinomatous metastases of the inguinal, iliac, peri-aortic abdominal lymph nodes, and of the liver, lungs, vagina and urinary bladder; extensive ulceration of the carcinoma of the rectum; fistulous passages from the rectum into the pelvic connective tissues and vagina; stricture at the rectosigmoid juncture; acute, minimal aspiration bronchopneumonia; old colostomy; old thrombophlebitis of left external iliac vein; acute, thrombo-ulcerative mitral endocarditis; slight hyperplasia of the spleen; anemia; emaciation; extensive fibrous pelvic peritonitis; cholelithiasis; left hydro-ureter and hydronephrosis; slight fatty changes of the aorta; accessory spleen."

Case 4.—The second patient, H., white, female, married, age 32, was admitted to the Cook County Hospital, November 30, 1925. Six years previously she had been operated upon for hemorrhoids and fistula-in-ano. Since then she had been troubled with recurrent ischiorectal abscesses and discharging fistulae. Examination showed a tight stricture of the rectum about 5 cm. from the anus and a marked induration of the rectum as far as it could be palpated through the posterior vaginal wall. Numerous rectovaginal and perirectal fistulae were present (Fig. 5). The patient was emaciated, anemic, and suffering from constant discharge of bloody pus from the rectum.

On December 21, 1925, an attempt was made to establish a left inguinal colostomy but the sigmoid and descending colon were found to be indurated and hyperemic, resulting in a marked preshortening of the mesentery of the sigmoid. Consequently, a cecostomy was performed. The patient was not seen again for two years, when she entered the hospital complaining of distention, diarrhea and abdominal pain. The rectum was almost completely occluded and the perirectal fistulae were still present. An ileostomy was performed at this time because of failure of the cecostomy to divert the fecal stream.

On September 27, 1932, seven years later, she returned saying that for five years she had been having attacks of unconsciousness and convulsions which were increasing in frequency. The first attack came on suddenly and without warning. She was taken to the hospital where she remained in a comatose state for four days. The diagnosis was idiopathic epilepsy. The epileptic episodes increased in frequency until February, 1933, at which time she was having from four to six attacks a week, each one lasting about two hours. Frei tests, given with several antigens, were all definitely positive. Knowing that leptomeningitis due to lymphogranuloma inguinale was possible, the patient was energetically treated with antimony and potassium tartrate, the dosage being 10 cc. of 1 per cent solution given intravenously. Later, Fuadin 5 cc. was given subcutaneously twice a week. From the very beginning of the treatment she improved, the attacks became infrequent and less severe, there having occurred only one seizure in five weeks, which lasted for ten minutes.

FIG. 5.—Case 4. Biopsy of lymphogranuloma inguinale of the rectum.

With ups and downs, this situation continued about the same until September, 1937, five years later, when the patient entered the County Hospital with increased pain in the rectum, induration of the rectovaginal septum, and increased discharge. The indurated area had broken down, a large crater ulcer developed, and a biopsy showed squamous cell carcinoma. The patient died on the tenth day after entering the hospital and an autopsy was performed with the following anatomic diagnosis:

Autopsy.—Anatomic Diagnosis: Ulcerated squamous carcinoma of the rectum engrafted upon a venereal proctitis and periproctitis, with extensive sclerosis of the perirectal fat (Figs. 6 and 7); carcinomatous metastases to the right inguinal lymph nodes; suppurative cystitis; slight bilateral hydronephrosis; cecostomy and ileostomy wounds; gangrenous abscess of the left upper lobe of the lung; fibroplastic deformity of the mitral valve with marked stenosis of the ostium; dilatation of the left cardiac auricle; cholelithiasis; subacute inflammatory hyperplasia of the spleen; cloudy swelling and fatty degeneration of the liver; fibrous adhesions about the uterus and adnexa, the liver, spleen and both lungs. The brain and meninges were negative.

In looking through the recent literature, two reports of lymphogranuloma inguinale involving the rectum complicated by the development of squamous cell carcinoma have been found: James R. Lisa (Arch. Path., **21**, 252, 1936; Transactions of N. Y. Path. Soc., November 26, 1935) reported the case of a Negro, age 37, with discharging perineal fistulae, a stricture of the rectum and a positive Frei test, negative Wassermann and Kahn. The process had

been present for ten years. Inguinal and femoral nodes were enlarged. Autopsy revealed a large adenocarcinoma of the pelvic colon with metastases to the iliac nodes and liver, and a squamous cell carcinoma of the anus with metastases to the left femoral nodes. The clinical diagnosis had been lymphogranuloma inguinale.

Bernstein (Am. Jour. Obst. & Gynec., 29, 718, 1935) reported the case of a Negress with extensive involvement of the anus and rectovaginal septum resulting from lymphogranuloma inguinale. Frei test positive. A squamous cell carcinoma developed in the vaginal fornix. Autopsy revealed the rectal tissue still showing typical evidences of lymphogranuloma inguinale.

FIG. 6.—Case 4. Gross specimen of squamous cell carcinoma developing on lymphogranuloma inguinale. Upper pointer in fistula to vagina. Lower pointer to fistula into the bladder.

FIG. 7.—Case 4. Biopsy of tumor arising from lymphogranuloma inguinale of the rectum.

These cases are suggestive as to the effect of long standing inflammation resulting from lymphogranuloma inguinale on the development of carcinoma.

Because of the disabling, serious clinical aspect of lymphogranuloma inguinale, as well as the tendency of squamous cell carcinoma to develop in the lesions, strenuous treatment should be instituted.

We have recently reported some favorable results in the treatment of the rectal lesions of lymphogranuloma inguinale with sulfanilamide. Whether or not that drug is specific has yet to be demonstrated, but in several cases with serious and widespread lesions the results were unbelievably good. In view of the tendency for squamous cell carcinoma to develop in long standing lesions of lymphogranuloma inguinale, we strongly recommend the use of sulfanilamide early in the course of the disease.

DISCUSSION.—DR. CURTICE ROSSER (Dallas, Tex.): The general subject which Doctor David has introduced, namely, the possible relationship between certain preceding irritative and inflammatory benign lesions of the anal canal, with cancer of the canal, is one in which I have been interested for several years. When, in 1930, I reported to the Section on Gastro-Enterology and Proctology of the American Medical Association that our experience indicated an apparent connection between certain lesions of the anal canal, particularly fistula-in-ano, and carcinomata which occur on that site later, it was with some temerity, because the literature then, as well as now, reflected considerable skepticism on this question. A statement made by Paul Kraske in the last century, that in his opinion there was no evidence to indicate that malignancy arises in hemorrhoids, fistulae or cicatrices, which has been copied from one text to another, is no doubt responsible for much of the opposition which exists to accepting a fact which now appears to us self-evident.

We have now observed a total of 16 cases of malignancy in the anal canal apparently induced by fistulae, hemorrhoids, ulcers and similar chronic lesions. It is interesting that in nine of these cases the malignancy itself has not been squamous cell cancer but adenocarcinoma, often growing in and under the skin of the anus. Two explanations may be offered for this fact: at the dentate line an overlapping of squamous and columnar cell epithelium occurs and malignant degeneration may arise in either type of tissue; moreover, as Hermann demonstrated in 1880, and as Tucker and Hellwig have emphasized recently, there are aberrant glandular structures lying under the anal skin which may also induce glandular cancer.

Lymphogranuloma venerea is a common rectal lesion in the South, especially among Negro women. Because it does produce a chronic irritative and inflammatory reaction, it had been a matter of considerable surprise to me that malignancy does not occur commonly from this disease. Since, however, we have observed in the teaching clinic of Baylor University School of Medicine over 200 cases, which have been followed during a long period of years, and have never detected a squamous cell cancer developing on one of the lesions, I am, therefore, quite unwilling to accept Doctor David's statement that the sequence he describes is of usual occurrence. My own opinion is that, in the cases he reports, the malignancy which developed arose from fistulae which were secondary to the lymphogranulomatous lesion.

DR. ALBERT O. SINGLETON (Galveston, Tex.): Doctor David reported two cases of squamous cell carcinoma of the rectum secondary to lymphogranuloma inguinale. Those of us who live in the South have naturally had experience in lymphogranuloma inguinale, because of its prevalence in the Negro race. We have often wondered why more of these have not resulted in malignancy. In examining our records of a series of cases we have had since 1920, we find we have had 181 clinically proven cases of lymphogranuloma with rectal involvement. The ages are not significant, except that it occurs largely in the age group from 30 to 50. In this entire group not one has developed carcinoma. One explanation may be that the patients do not live very long; comparatively few have reached the age of 60. This short duration of life may account for the absence of carcinoma. There were 14 males, 177 females; eight were white, 173 Negroes. The Wassermann test was positive 57 times, negative 98, and unknown 26. The Frei test has been used in all suspected cases since 1934. In the same period we have had 47 adenocarcinomata of the rectum and seven epitheliomata or flat cell carcinomata. None were preceded by lymphogranuloma inguinale. The fact that pigmented skin does not predispose to carcinoma so readily as the white skin may be a factor.

INTESTINAL OBSTRUCTION DUE TO INTRALUMINAL FOREIGN BODIES

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VARIOUS aspects of the problem of intestinal obstruction due to intraluminal foreign bodies have previously interested members of the Southern Surgical Association. Dr. Frank Martin,[1] of Baltimore, at the 1911 meeting of the Association in Washington, D. C., presented a paper on "Intestinal Obstruction Due to Gallstones," and Dr. M. J. Henry,[2] of Louisville, in 1931, presented a paper before this society on the same subject at the meeting in White Sulphur Springs, West Virginia. Dr. Webb Griffith,[3] in 1935, in Hot Springs, Virginia, discussed "Obstruction of the Small Intestine Due to Food Products." Dr. Fred Rankin[4] has written on the subject of gallstone ileus, and Dr. Alexius McGlannan[5] has described the occurrence of intussusception in association with round worm infestation. Doctors Crisler[6] and Frank[7] have each reported cases of intestinal obstruction due to food substances. The development and manifestations of intestinal trichobezoars have been studied and reported on by Matas.[8]

The occult character of intestinal obstruction due to intraluminal foreign bodies and the consequent delay in the institution of therapy are responsible for a high mortality in this type of ileus. Whereas, the usual absence of an obvious cause is common to all types of intraluminal foreign body obstruction, the manifestations and management of the many types within this general group vary widely and depend upon the specific etiology. In a series of 875 Charity Hospital cases of intestinal obstruction, which did not include obstruction due to herniae, neoplasms, or peritonitis, there were 51 instances of intestinal obstruction due to intraluminal foreign bodies, an incidence of 5.8 per cent (Chart 1).

The following outline indicates the classification of causes of intraluminal foreign body obstruction which we believe clearly differentiates the types of ileus to be considered:

I. Gallstones.

II. Intestinal parasites: (1) Ascaris lumbricoides. (2) Taenia. (3) Trichocephalus. (4) Oxyuris.

III. Fecaliths (strecoliths, coproliths).

IV. Enteroliths: (1) Cholesterol and choleic acid. (2) Calcium carbonate and phosphate.

V. Concretions: (1) Shellac. (2) Casein. (3) Raw latex (caoutchouc). (4) Bismuth and magnesium carbonate. (5) Miscellaneous concretions.

VI. Bezoars: (1) Phytobezoars: (a) Diosporobezoars (persimmon);

(b) miscellaneous phytobezoars. (2) Trichobezoars. (3) Miscellaneous bezoars.

VII. Food boli: (1) Legumes. (2) Fruits. (3) Oats. (4) Vegetables. (5) Meat. (6) Seeds. (7) Grasshoppers, *etc.*

VIII. Miscellaneous foreign bodies: (1) Metal. (2) Glass. (3) Bone. (4) Murphy buttons, *etc.*

IX. Meconium ileus and epithelial casts.

Because of the similarity in gross appearance of gallstones, fecaliths, enteroliths, and intestinal concretions, as well as some types of bezoars, especially diosporobezoars, considerable confusion concerning the proper classifica-

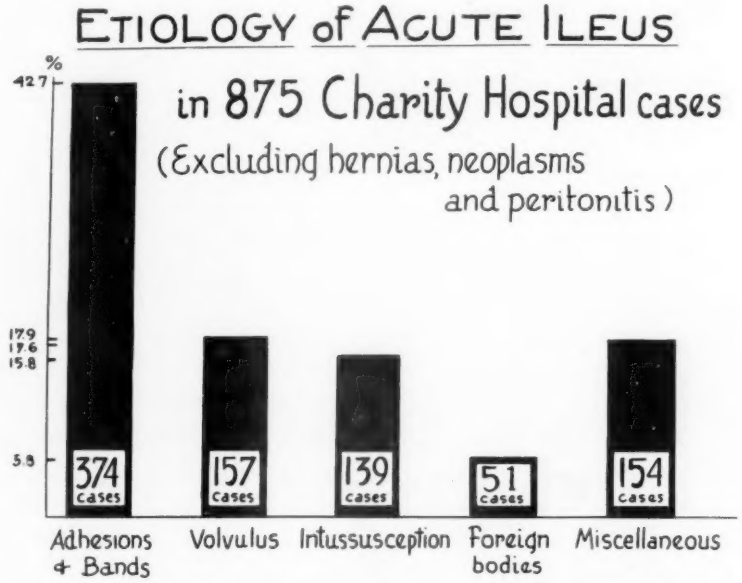

CHART 1.—Graphic representation showing the relative frequency of intestinal obstruction due to intraluminal foreign bodies in the authors' series. This type of ileus occurred in 5.8 per cent of the cases.

tion of these intraluminal foreign bodies is found in the literature. This is particularly true when careful chemical or microscopic examinations of the specimens has not been made. In the present classification, by the term "gallstone" we mean those true cholecystoliths which are formed within the gallbladder and which, subsequently, find their way into the intestinal tract via cholecystoduodenal or cholecystocolonic fistulae. Comprehensive reviews of gallstone ileus have been published by Waring,[9] Treves,[10] Robson,[11] Moynihan,[12] Murphy,[13] Martin,[1] Brown,[14] Wagner,[15] Jones,[16] Burgess,[17] Rankin,[4] Judd,[18] Odess,[19] Powers,[20] Folliasson,[21] Duboucher,[22] Henry,[2] Dickson,[23] Gasper,[24] and Balch[25] (Fig. 1). We have used the term "fecalith" to indicate inspissated masses of feces (Treves,[10] Downing,[26] Fischer,[27] and Bottino[28] (Fig. 2).

The term "enterolith" is applied to those intestinal calculi of definite chemical composition which have formed primarily within the intestinal lumen.

The chemical composition of some of the enteroliths rather closely corresponds to the chemical composition of certain gallstones, whereas other enteroliths have a composition resembling, or at least likely to be confused with, medica-

FIG. 1.—Photograph of a gallstone which caused obstruction in one of the authors' series. In this case enterotomy was performed, with closure of the intestine with interrupted silk. The patient made an uneventful recovery.

FIG. 2.—Photograph showing fecalith removed in one of the authors' cases. Cross-section of this lith showed distinct lamination which can be seen in Figure 5, which is a roentgenogram showing the same specimen *in situ*.

ment concretions. By the term "concretion" is meant a foreign body which is homogenously composed of ingested foreign substances or chemicals. Winterstein,[29] Lichtenstern,[30] Friedländer,[31] deLangenhagen,[32] Treves,[10] Halstead,[33]

Matignon,[34] DeBakey and Ochsner,[35] and Nothnagel[36] have reported on enteroliths and concretions. "Bezoar" is used to indicate those intraluminal foreign bodies which are made up of agglutinated or intertwined organic substances. A comprehensive review of the literature on bezoars with case reports

FIG. 3.—Photograph showing external appearance of a persimmon phytobezoar, which caused intestinal obstruction in one of the authors' series. A persimmon seed, probably the nucleus of this mass, is indicated.

FIG. 4.—Drawing showing impacted mass of intertwined intestinal parasites (ascaris lumbricoides), which caused intestinal obstruction.

has very recently been completed by DeBakey and Ochsner.[35] Butterworth[37] has made a special study of the occurrence of bezoars in children, and Matas[8] has written concerning hair casts of the intestinal tract (Fig. 3). Intestinal obstruction produced by food boli has attracted the attention of Tigi,[38] Brizke,[39]

Elliott,[40] Caleveart,[41] Griffith,[3] Crisler,[6] Frank,[7] Caylor and Nickel,[42] and Leisinger.[43] The rôle played by intestinal parasites, as a cause of ileus, has been recorded by Davaine,[44] Fevre,[45] Rios,[46] Faust,[47] McGlannan,[5] Hall,[48] Watson,[49] Brown,[50] Leitch,[51] and Greene[52] (Fig. 4). Intestinal obstruction due to inspissated meconium or to epithelial casts has been reported as a cause of ileus in the newborn, and resembles intestinal obstruction caused by the impaction of feces in adults. Meconium ileus in several instances has been associated with stenosis of the pancreatic duct, thus accounting for the suggestion by Kornblith and Otani,[53] Landsteiner,[54] and Farber[55] that the development of this condition is due to a deficiency of pancreatic secretion. Exalto[56] and Fanconi,[57] on the other hand, believe that the accumulation of a cement-like mass of meconium is, at least sometimes, due to a disturbance in the bile

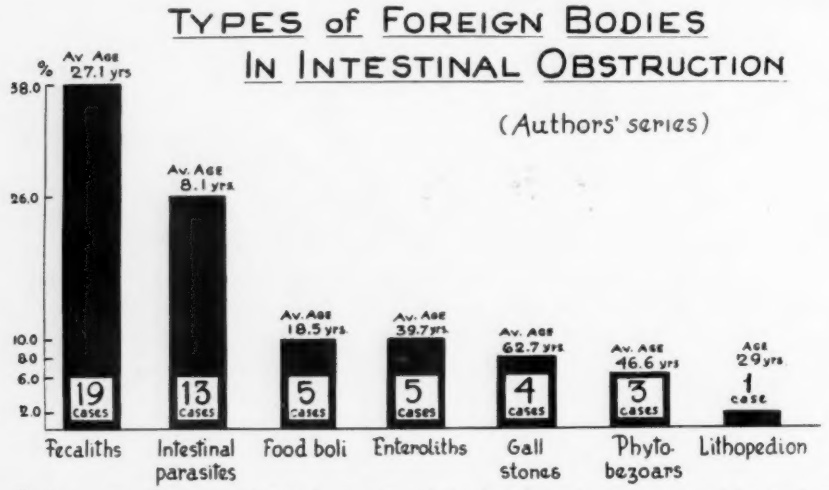

CHART 2.—Graphic representation showing the relative incidence of various types of foreign bodies causing intestinal obstruction in the authors' series. The apparent frequency with which fecaliths were the obstructing agents is, no doubt, in part due to the erroneous designation of other liths, not subjected to careful chemical or microscopic examination, as fecaliths.

secretion. Miscellaneous agents in addition to those already considered may cause intraluminal obstruction. In the authors' series, for example, intraluminal obstruction was in one instance due to a lithopedion which eroded the rectal wall and obstructed the bowel lumen.

The incidence of the various types of intraluminal foreign body obstruction and the average age of the patients in each group in the authors' series are shown in Chart 2. The apparently high incidence of fecaliths as a cause of obstruction in the cases which we have collected may in part be due to the fact that the specimens removed were in some instances not subjected to microscopic examination or chemical analysis, and it is entirely possible that some of these liths were gallstones, enteroliths, concretions, or even phytobezoars. The relatively high incidence of intestinal parasite infestation in the New Orleans area accounts for intestinal parasites being the second most frequent cause of intraluminal ileus in the authors' series. The occurrence of this type

of obstruction more frequently in young individuals is probably related to the smaller caliber of the intestine as well as to the more common existence of intestinal parasite infestation in children. Although a variety of intestinal parasites may cause obstruction, a vast majority of the reports on this type of ileus concern cases of ascaris lumbricoides infestation, and in our own series all the obstructions of this sort were due to ascarids. The comparative frequency of obstruction due to ascarids is evidently due not only to the widespread distribution of this parasite and the common occurrence of this type of infestation, but to the size and shape of the parasites as well as the great number of such worms which often are present in an infested individual. Although there were, in our series, an equal number of cases of obstruction due to food boli and to enteroliths, there was a striking difference in the average age of the patients in these two groups. In the food bolus obstruction group, the average age was less than one-half that of the enterolith group. The food bolus obstructions were in two instances due to masses of watermelon seeds, and in one instance each, masses of peanut particles, raw butter beans, and raw sweet potato were the causative agents. The age of the gallstone ileus cases corresponds with the reports of others, and accounts for a relatively high mortality in this group. The incidence of obstruction due to phytobezoars appears to have been relatively low, but we believe that at least in some of the cases in which the obstructing agents were described as fecaliths, the foreign bodies were actually phytobezoars. There were no cases of ileus due to trichobezoars among the cases which we collected.

The possibility of intestinal obstruction being caused by a Murphy button was a matter of concern when such devices were more frequently used, and there have been reports on the occurrence of intestinal obstruction following the employment of such anastomosis apparatus (Beer,[58] Gelpke,[59] and Kloiber[60]). Eve's[61] report on "The Case of the Human Ostrich" is a classic example of intestinal obstruction caused by the ingestion of miscellaneous foreign bodies. In none of our collected cases could the obstructing agent be designated as a true intestinal concretion, according to the definition of an intestinal concretion which we have used. The Charity Hospital records failed to reveal any cases of intestinal obstruction due to meconium or to epithelial casts. Wangensteen,[62] in a recent monograph, has reviewed the occurrence as well as the therapeutic problems in the management of foreign body ileus.

The development of intraluminal foreign body obstruction is usually dependent upon definite predisposing elements or circumstances. Such factors as climate, geographic location, and hygienic conditions are intimately related to the occurrence of ileus due to intestinal parasites. Occupation may be a predisposing cause, as in the instance of furniture workers, who form concretions as the result of drinking shellac for its alcoholic content. Occupation is also related to the development of concretions of raw latex or caoutchouc among workers with this substance. Economic conditions which influence food supply may be of paramount importance as exemplified by the cases of obstruction which occurred during the World War when oats were rationed

as food in Russa (Brizke[39]), and when "ammunition bread" was resorted to in Germany. The custom of eating the grasshoppers (Caleveart[41]) which have decimated crops in the Belgian Congo has likewise been responsible for the development of intestinal obstruction.

Congenital or acquired stenosis of the intestinal lumen as well as kinking or acute angulation of the intestine may serve as predisposing causes of intraluminal obstruction. Digestive disturbances such as deficient digestive juices may be responsible for incomplete disintegration of food with resulting formation of an obstructing bolus. Constipation, with associated excessive dehydration and inspissation of feces, may act as a predisposing cause in the development of fecalith obstruction. Even psychologic aberrations, such as those responsible for the habit of hair swallowing, with resulting trichobezoar formation, may lead to intraluminal obstruction.

The exciting causes of intraluminal obstruction include improper eating habits such as insufficient mastication by the hurried adult, food-bolting by children, eating without benefit of normal or artificial teeth, dilution of digestive secretions by drinking excessive quantities of water with meals, and overloading of the intestinal tract with unusually large amounts of food. The character of the food is significant in the development of food bolus obstruction, as such obstructions almost invariably occur following the ingestion of desiccated food, such as dried fruits, or fiber-rich food. The character of the foodstuff is also important in the instance of persimmons, the skins of which contain shibuol, a cement substance which, when precipitated by hydrochloric acid in the stomach, holds together the persimmon seeds, fibers, and pieces of skin, with consequent formation of a bezoar, all or part of which may pass into the intestine and cause obstruction. The accumulation of excessive amounts of gas in the intestine, resulting from fermentation following the eating of beans, has been cited as an exciting cause of intestinal obstruction, as the excessive distention of the intestine may result in angulation or kinking. The administration of a vermifuge can precipitate intestinal obstruction in individuals with a heavy infestation of intestinal parasites. Either the massing together of large numbers of intestinal parasites or the intense contraction of the intestine at the site of a single parasite may produce mechanical obstruction. The liberation of toxic substances from dead and decomposing worms may cause an adynamic ileus, systemic intoxication and even death.

The pathologic changes observed in intestinal obstruction due to intraluminal foreign bodies are the same as those which are usually present in ileus due to other causes. Intussusception may occur as a result of intestinal spasm associated with hyperperistalsis and thus cause complete mechanical obstruction in individuals harboring intestinal parasites. Intramural vascular strangulation may be produced by excessive distention of the intestine. Local pressure necrosis is particularly likely to occur in the presence of hard unyielding gallstones, enteroliths, fecaliths, intestinal calculi, hard bezoars, and concretions; and unless relieved, necrosis or gangrene of a segment of intestine may occur.

Perforation at a site of lowered tissue resistance has been observed in some instances of intestinal parasite infestation associated with intestinal obstruction. Many cases have been reported in which incarceration or strangulation of an external hernia has been precipitated and then maintained by the accumulation of intestinal parasites within the segment of bowel located in a hernial sac.

Of paramount importance in the recognition of intraluminal foreign body obstruction is careful and special history-taking. There may or may not be a history of pelvic or generalized peritonitis, suggesting that obstruction may be due to bands or adhesions. Such adhesions may, indeed, constitute the

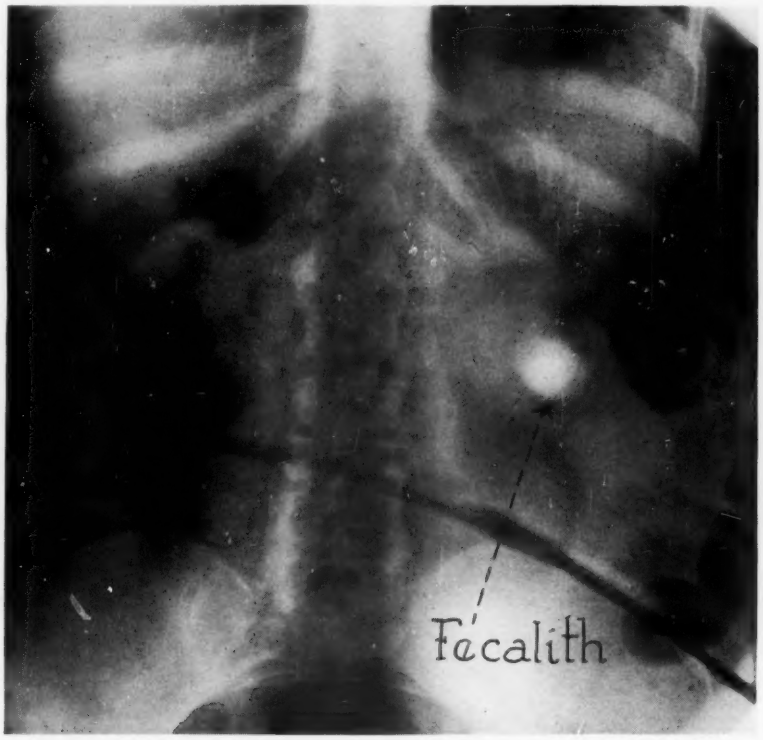

FIG. 5.—Roentgenogram showing fecalith and distended loops of small bowel in which valvulae conniventes are distinctly outlined.

background for an intraluminal obstruction by causing kinking or angulation of the intestine. In order to arrive at a complete and accurate diagnosis, detailed information concerning such factors as eating habits, evidences of intestinal parasites, and manifestations or gallbladder disease must be sought. The clinical picture presented by the patient in whom intestinal obstruction is due to an intraluminal foreign body is essentially the same as that observed in cases of intestinal obstruction from other causes. The absence of an obvious cause of the ileus frequently leads to delay in making the diagnosis and to dangerously long periods of observation. The physical findings in this class of cases are essentially the same as those which result from intestinal

obstruction due to other mechanical causes. The palpation of a tumor may at times be possible, and in the instance of obstruction due to intestinal parasites some observers have reported the palpation of a verminous mass or even the detection of slight movements due to the activity of the worms. When intussusception occurs, the characteristic sausage-like mass may be felt; and when volvulus of the large intestine is associated with the intraluminal obstruction, eccentric distention of the abdomen may be observed.

The roentgenologic findings in intraluminal obstruction consist of the same abnormal accumulations of gas or fluid observed in other types of mechanical ileus. The possibility of occasionally visualizing the foreign body roentgenographically makes it advisable to have a plain roentgenogram of the abdomen of all cases suspected of having intestinal obstruction due to an intraluminal foreign body (Fig. 5).

Differential diagnosis in cases of intestinal obstruction due to intraluminal foreign bodies is frequently difficult. Depending upon the acuteness or the chronicity of the symptoms it may, for instance, be necessary to differentiate this condition from acute gastro-enteritis. Acute appendicitis and other acute

TABLE I

PREOPERATIVE COMPLICATIONS AND ASSOCIATED CONDITIONS

Present in eight of 50 cases—16 per cent

	Authors' Series Number of Cases
Perforation and peritonitis	2
Internal hernia	2
Inguinal hernia	1
Peptic ulcer	1
Stenosis following regional enteritis	1
Phytobezoar in stomach	1

surgical conditions may be confused with intraluminal foreign body obstruction. Furthermore, the presence of a mass, especially when there is a history of antecedent partial intestinal obstruction, has led to an erroneous diagnosis of intestinal neoplasia.

Preoperative complications and associated conditions which have already been referred to under the discussion of pathology were present in eight of the authors' 50 cases, an incidence of 16 per cent. The distribution of these complications and associated conditions is indicated in Table I.

Perforation of the intestine, with resulting peritonitis, occurred in three of the cases we are reporting, and has been frequently cited as a complication of obstruction due to the hard foreign bodies, notably fecaliths, enteroliths, concretions, phytobezoars, and gallstones. This complication is associated with an extremely high mortality, and two of the three cases in our series in which it developed, ended fatally. In one of these cases, the perforation was caused by a gallstone; a persimmon phytobezoar was responsible for the small perforation in another case; and in still another case an impacted enterolith eroded the intestinal wall (Figs. 6 and 7).

In two of the cases an internal hernia existed in conjunction with the intraluminal obstruction. The likelihood of this latter complication developing in association with intraluminal obstruction is heightened by distention of the intestine, which may also be responsible for secondary "water-hose kink"

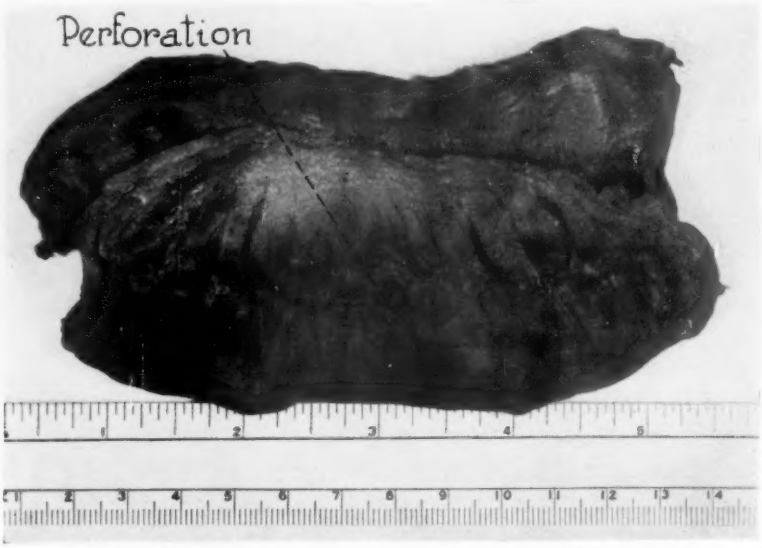

FIG. 6.—Photograph of a segment of intestine showing the point of perforation, due to pressure necrosis produced by a persimmon phytobezoar.

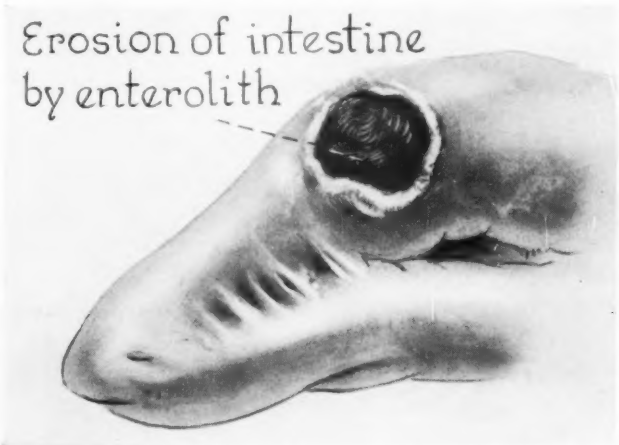

FIG. 7.—Drawing showing erosion of the intestinal wall by an enterolith lodged above the site of an intestinal angulation caused by adhesions.

obstruction. Of the two cases in which an internal hernia existed, one died and one lived. In one instance recurrent intraluminal obstruction due to a fecalith was associated with an inguinal hernia, the intraluminal obstruction due to the fecalith occurring in the herniated segment of intestine. This patient survived. A coexisting peptic ulcer complicated one of our cases. A

residual stenosis following regional enteritis served as a predisposing factor in one of the intestinal parasite obstruction cases. A diosporobezoar was present in the stomach of one of our collected group of intestinal obstructions due to phytobezoars. It could not be determined whether the intestinal phytobezoar had been formed independently or if it was a detached portion of the gastric bezoar.

Therapy in intraluminal ileus may be nonoperative or operative. Nonoperative therapy may be further subdivided into conservative, radical, and palliative. By conservative nonoperative treatment we mean observation of the patient and administration of general supportive measures such as venoclysis, the application of heat to the abdomen, the withholding of food by mouth, the institution of gastroduodenal suction, the administration of antispasmodics, notably atropine and belladonna, and occasionally the employment of enemata. Such a regimen of treatment, especially in the instance of food bolus and intestinal parasite obstructions, is frequently adequate. The objection to the conservative method of treatment, however, is that it is likely to be persisted in for an unduly long time, and many of the bad results credited to the operative method of treatment are due to the unwarranted persistence in the application of conservative methods. The radical nonoperative treatment, which consists of the administration of purgatives and vermifuges without knowing the extent and character of the obstruction, we consider to be dangerous. Palliative conservative treatment may be resorted to in the instance of extremely ill patients, at least during the period when the election of a more radical type of treatment is out of the question.

Operative treatment may also be subdivided into conservative, radical, and palliative procedures. Conservative operative treatment consists of exploratory celiotomy for the purpose of determining the location, extent, and general character of the obstruction, and this sometimes may be supplemented by dissociation or displacement of the obstructing mass. The advantage of this latter type of therapy, *i.e.*, avoidance of opening the intestine while at the same time affording relief of impaction, is reflected in the relatively low mortality following its employment. Radical therapy may consist of enterotomy, enterostomy, exteriorization, stripping out the contents of the intestine, or resection. Of these various operations, enterotomy appears to be the ideal procedure. The fixation of a loop of intestine to the abdominal wall and the attendant danger of secondary intestinal obstruction constitute a definite objection to the performance of enterostomy. Exteriorization of a loop may be necessary as a life-saving procedure in the advanced cases in which damage of the intestine has occurred. Resection is rarely indicated and is to be avoided in all except the extreme cases in which this procedure is imperative. Stripping or milking-out the contents of the obstructed bowel, although occasionally necessary in order to permit reposition of the intestine following accidental or intentional evisceration, is another procedure which should usually be avoided. Observations by Läwen[63] and ourselves[64, 65] have shown that this latter procedure causes an increase in the degree of adynamic ileus, and the shock associated

with this operation is likely to lead to death due to a persistent lowering of the blood pressure. Palliative enterostomy may be the procedure of choice when it is impossible to locate the cause of the obstruction or when no more extensive procedure seems warranted.

The nature and ability to recognize the causative agent, the presence or absence of specific complications, the general condition of the patient, and the time elapsed since onset of symptoms determine the mode of treatment. Based upon the foregoing considerations, the following is offered as a guide in selecting the type of management to be employed in cases of intestinal obstruction due to intraluminal foreign bodies, especially when the exact cause of the obstruction is not known:

I. Indications for Conservative Treatment or Delay in Institution of Surgical Treatment:

A. Early cases.
(1) Very short interval since onset of symptoms.
(a) Absence of significant or definite roetgenologic findings.
(2) Indefinite evidence of mechanical obstruction and especially the absence of findings suggesting intussusception, volvulus, or mesenteric thrombosis.

B. Moderately advanced cases.
(1) No indications for delay except extremely rapid and complete relief following a short period of conservative treatment. This applies in particular to elderly, extremely obese, or otherwise poor-risk patients.

C. Advanced cases with pronounced dehydration and distention in which preoperative preparation is imperative should, at least, receive preliminary conservative treatment. In many of these cases a totally conservative management offers the only likely chance of survival.

II. Indications for Early or Immediate Operative Intervention:

A. Clinically indefinite, early or moderately advanced cases with, however, roentgenographically demonstrable calculus, enterolith, gallstone, or fecalith.

B. Early cases with definite symptoms or signs of obstruction with or without significant roentgenographic findings such as abnormal gas accumulations or fluid levels.

C. Questionable or borderline cases in which symptoms or signs suggestive of intestinal obstruction persist following a short period of nonoperative treatment.

D. Moderately advanced cases with evidence of intussusception, volvulus, or mesenteric thrombosis.

E. Palpation of an abnormal mass.

F. Evidence of perforation.

When the specific type of foreign body is known or suspected, elective therapy may be considered in relation to the consistency of the foreign body.

The nonresilient foreign bodies, such as gallstones, fecaliths, enteroliths, concretions, and the bezoars of hard consistency which, because of their chemical composition or physical properties, will not spontaneously change their form and which are likely to become progressively more firmly impacted, require

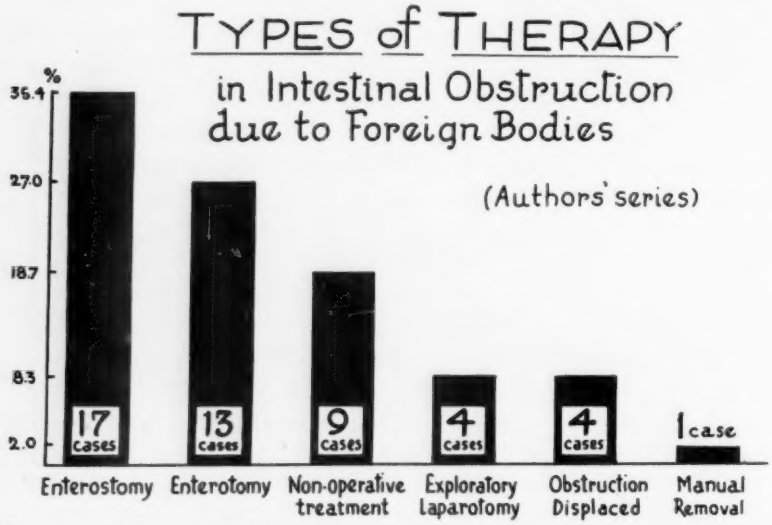

CHART 3.—Graphic representation indicating the types of therapy employed in the management of intestinal obstruction due to foreign bodies in the authors' series.

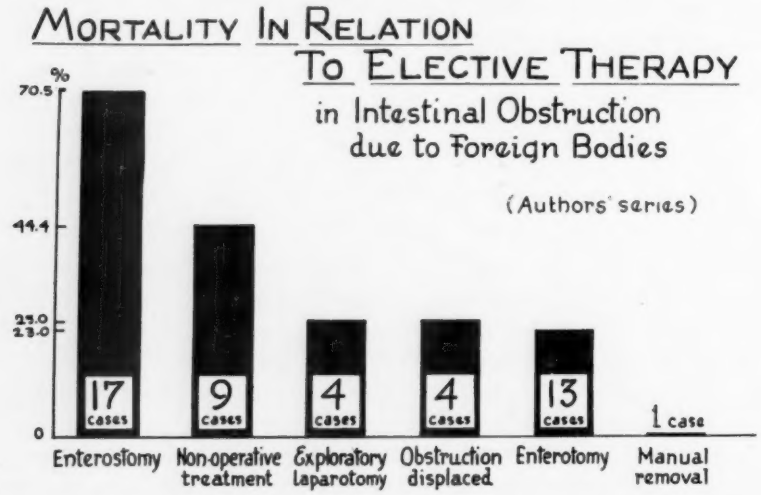

CHART 4.—Graphic representation showing the mortality in relation to elective therapy employed in the management of intestinal obstruction due to foreign bodies in the authors' series. The most frequently performed procedure, namely, enterostomy, was accompanied by the highest mortality. The low mortality, associated with exploratory celiotomy and with displacement or dissociation of the obstructing agent, indicates that these procedures may be advantageously employed. The comparatively low mortality following enterotomy, indicates that this is the better of the radical operative methods of treatment.

early operative removal. If irregular in contour or presenting angulated surfaces, these foreign bodies are particularly likely to cause erosion of the intestinal wall. On the other hand, the relatively soft, compressible, or

malleable, foreign bodies such as food boli and intestinal parasites are not likely to cause erosion of the bowel. Food boli may disintegrate as a result of the action of digestive juices or bacteria, causing softening or liquefaction of the mass, and intestinal parasites may migrate and thereby relieve the obstruction. Therefore, patients with obstruction due to these two latter causes frequently may be kept under observation or treated conservatively. Even when operative intervention seems indicated or when operative exploration has been performed without preoperative suspicion of the presence of these foreign bodies, the more conservative operative procedures may be employed.

The various types of therapy employed in the authors' series are indicated in Chart 3. In 17 of the Charity Hospital cases, enterostomy was performed. The frequency with which this procedure was employed was, presumably, due to the considerable number of instances in which conditions such as extreme intestinal distention made this operation necessary. Enterotomy was performed in 13 cases and is, we believe, the operation of choice in those cases in which it is necessary to open the intestinal lumen. An entirely nonoperative method of treatment consisting of the administration of antispasmodics, enemata, and vermifuges was employed in nine cases. Exploratory celiotomy alone was performed in four cases, while in another four cases in addition to the exploratory celiotomy, the foreign body was displaced. Manual removal of the obstructing agent per rectum was performed in one case.

The mortality in relation to the elective therapy in the authors' collected cases is graphically represented in Chart 4. The most frequently performed operative procedure, enterostomy, was associated with the highest mortality. In the 17 cases in which it was performed, there was a mortality of 70.5 per cent. The next highest mortality group was the one in which nonoperative treatment was employed. In the nine cases in which this type of therapy was used there was a mortality of 44.4 per cent, due in part to the fact that this method of management was more or less necessarily followed in several cases in which the patient's extremely poor condition precluded more radical methods of treatment. The mortality following either exploratory celiotomy or displacement of the obstruction was equal, being in each instance 25 per cent. Of the 13 cases in our series in which enterotomy was performed, 23 per cent died. The fatality rate in the group in which enterotomy was performed was only one-third as great as in the group in which enterostomy was performed. However, the group in which the latter procedure was employed included many cases of advanced obstruction. The one case in which manual removal of an obstructing fecal mass was done, lived.

SUMMARY

Intraluminal obstruction due to foreign bodies is not uncommon. This type of ileus may be produced by a variety of obstructing agents including gallstones, intestinal parasites, fecaliths, enteroliths, intestinal concretions,

bezoars, food boli, inspissated meconium, epithelial casts, and miscellaneous foreign bodies.

The authors have reviewed 875 cases of intestinal obstruction, which did not include intestinal obstruction due to herniae, neoplasms, or peritonitis. There were 51 instances of intestinal obstruction due to intraluminal foreign bodies, an incidence of 5.8 per cent.

In the authors' series, fecaliths were the most common causes of intraluminal intestinal obstruction, and the next most common causative agent was intestinal parasites.

Factors which influence the development of intraluminal foreign body obstruction include climate, geographic location, hygienic conditions, occupation, food supply, and eating habits. Congenital or acquired stenosis of the intestinal lumen, as well as kinking or acute angulation of the intestine, predisposes to the establishment of intraluminal obstruction by foreign bodies.

Intussusception may be induced by intestinal parasites which cause intestinal spasm and associated hyperperistalsis.

Perforation of the intestine due to pressure necrosis is particularly likely to occur in the presence of hard foreign bodies such as gallstones, enteroliths, fecaliths, intestinal calculi, concretions, and nonresilient bezoars.

Careful history-taking is especially important in the diagnosis of intraluminal intestinal obstruction. There are usually no distinctive physical findings. Roentgenologic examination may reveal the obstructing agent.

Depending upon the severity of the symptoms and the duration of the obstruction, intraluminal ileus may be confused with acute gastro-enteritis, various acute surgical abdominal conditions, or even intra-abdominal neoplasia.

Treatment may be either nonoperative or operative, and each of these two general types of management may be further subdivided into conservative, radical, and palliative methods of therapy. The nonoperative method of treatment may be most advantageously employed in the instance of the soft or malleable foreign bodies, notably food boli and intestinal parasites, which are not likely to cause perforation. On the other hand, early operative treatment of the type herein described as radical is usually to be preferred in the instance of the hard or unyielding foreign bodies which may cause pressure necrosis of the intestinal wall with subsequent perforation and peritonitis. In some instances, operative exploration of the abdomen and displacement or dissociation of the obstructing mass without opening the intestinal lumen may adequately relieve the obstruction.

Of the radical operative measures, enterotomy is the procedure of choice. The present satisfactory methods of treating ileus make enterostomy rarely necessary.

Detailed criteria for the application of the various types of therapy are presented in this report.

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DISCUSSION.—DR. JOHN GERSTER (New York, N. Y.): Regarding ascarides, Flury's[1] exhaustive report, in 1912, upon their irritating poisons and his reference to Peiper's[2] article on the many different poisons from all the varieties of intestinal parasitic worms are of interest, not only because of intestinal obstruction but also because of the other manifold local and general symptoms arising from absorption of their emanations.[3]

I should like to report the case of a man, age 60, with a distended abdomen of five days' standing, with obstruction, vomiting, tenderness, and it was impossible to make an exact diagnosis. On opening the abdomen in the midline, an enormously distended cecum, filled with fluid feces, was found, which immediately began to leak fluid stool through a pinhole perforation. The mobile cecum was promptly delivered so that peritoneal soiling was avoided. A cecostomy was immediately established. The patient then developed a pneumonia and delirium tremens from which, however, he recovered. After repeated colonic irrigations, a barium enema showed an obstruction of the sigmoid. The patient asked for a continuation of the daily irrigations "to see if the obstruction would go away." The irrigations were continued daily for another week. Finally, several round balls of hard dry fecal matter came away. The cecostomy was closed. He is well.

Regarding intestinal obstruction from eating grasshoppers, one might imagine that if John the Baptist had not added honey to his diet of locusts, a bolus might have killed him and so he would not have featured in the incidents with Salome and Herod.

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TRICHOBEZOARS

REPORT OF A CASE WITH RECURRENCE

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THE occurrence of a trichobezoar or hair ball in the human gastro-intestinal tract, while not rare, is infrequent enough to arouse interest and may well be considerad among the curiosities of medicine. The writings of the ancients reveal that, as early as the twelfth century B.C., it was commonly known that hair balls were frequently found in the stomach of animals, but it was not until 1779 that Baudamant,[1] in an autopsy report, recorded the first trichobezoar found in man. The patient was a boy, age 16, who was reputed to have been eating hair since infancy. He had diarrhea, frequent and violent vomiting and fever. He finally died of exhaustion. At autopsy two masses of hair were found—one in the stomach and the other in the jejunum. In 1883, Schönborn[2] reported the first operation for removal of a hair ball from the stomach of a human. The patient was a girl, age 15. The preoperative diagnosis rested between a floating kidney, movable spleen or a tumor of the omentum. After a stormy convalescence she made a complete recovery.

During the past half century, the literature of trichobezoar has been enriched by many important contributions, probably the most noteworthy of which was a paper presented by Dr. Rudolph Matas[14] before the Southern Surgical Association, in 1914. It was a complete and most instructive exposition of the subject. He had collected a total of 76 cases. Forty-seven of these had been found at operation and the remaining 29 had been found at autopsy. Maes,[21] in a review of the literature, in 1928, found that 40 additional cases had been reported since 1914, making a total in all of 116 cases. I find after a thorough search of the literature that since the publication of Maes' paper in 1928, there have been ten other cases reported from the United States,[22, 27, 33, 34, 35, 36, 37, 40] one from Canada,[32] four from Germany,[24, 26, 29, 38] two from Great Britain,[25, 39] two from Italy,[28, 30] one from Argentina,[31] and one from Brazil[23]—21 in all, which, in addition to the case herewith reported, makes a total of 138 cases. As Matas suggested, there are no doubt many unreported cases that have been operated upon, which, if reported, would swell the statistics considerably.

In studying the history of the cases that have been reported, an interesting fact was observed, namely, the small number of operations that have been performed for recurrences. Only three instances of recurrence appear in the literature. The first case of recurrence is that reported by Graeve[18]: A girl, age 6, who had been eating her own hair since she was four years old. A hair ball was removed, January 2, 1917, and on November 22, 1918, a second hair ball was removed. The second case, reported by Harris,[20] in 1925, is so unusual that I feel it merits description in detail—five hair balls having been

removed within 13 years. The following is quoted by permission of the author:

"In 1911, an unmarried woman, age 35, was operated upon . . . for the removal of a hair ball from her stomach. In April, 1920, she was operated upon . . . for similar condition, and again in June, 1922. When I saw her in October, 1923, she complained of pain and a swelling in her abdomen and loss of appetite of three months' duration. On examination a large solid tumor was visible and palpable in the upper part of the abdomen, and in view of her past medical history another hair ball was suspected. Upon opening the stomach, which was much enlarged, it was found to be entirely occupied by a mass of hair. This extended through the pylorus into the duodenum and when removed was found to weigh 2½ pounds. It was 20 inches long and its girth at its largest circumference in the stomach portion was 15 inches. Recovery was uneventful and the patient, after being given a solemn warning to refrain from the habit of hair eating, was discharged as cured. In November, 1924, she once more complained of the return of her former trouble, and I again removed a large hair ball. Up to the present I think she remains quite well. Apart from her hair-eating habits this woman is quite normal mentally, being well educated and a very fair musician. She seems to have suffered no permanent ill effects from her repeated operations since her abdominal wall is firm, and when her stomach is not full of hair her digestion is unimpaired."

The third recurrent trichobezoar was reported by Bennett,[34] in 1934. The patient, a female, age 50, had had an operation 20 years previously for the removal of a hair ball weighing two and one-half pounds At the second operation, in 1933, a hair ball weighing three and one-half pounds was removed.

Neely,[19] in 1924, reported that he had removed a hair ball from a girl, age 11, and that she began eating hair before she left the hospital. There is no record, however, that she was operated upon subsequently. There is no satisfactory explanation as to why recurrences have not been more frequent.

The cause of this strange perversion is obscure. Rovsing[17] stated, in 1924, that in his opinion trichobezoars occur only in the mentally feeble and insane. However, this opinion is not shared by other writers. On the other hand, the vast majority of these individuals are normal or above normal in mentality. It is true that some insane persons eat hair, but they also usually eat other more solid substances, such as nails and wire. To date, only six cases have been reported in males. The first was the historic case reported by Baudamant,[1] in 1779. The second was that reported by Cobbold,[4] in 1886—an idiotic boy, age 18, in whom a two and one-quarter pound hair ball was found at autopsy. The third case occurred in an insane man, age 43, reported by Gemmel,[5] in 1894. When his stomach was opened it was found to be filled with nails, wire and other foreign bodies, with a hair ball in his esophagus projecting into the stomach. The fourth case was reported by Costa and Tasca,[30] in 1932. The patient was a grief stricken widower who, shortly after the death of his wife, ate a large collection of her hair combings which she had been accumulating to sell. The fifth case was reported, in 1933, by Faulkner and Adams.[32] The patient was a man, age 31, of normal mentality. The sixth case was reported by Kerr and Rypins[33]—a man, age 20, who also had a normal mentality. Just why this perverted appetite should be so much more common in females than in males is difficult to understand. There is probably some unknown psychic or neurologic factor peculiar to females in the adolescent period of life which plays a large part in the etiology. The explanation offered by the older writers for the formation of these tumors was that women

and young girls wore long hair and frequently acquired the habit of passing strands of hair through the mouth and between the teeth, resulting in their biting off bits of hair and swallowing them. However, as Maes observes, since bobbing the hair has become the almost universal style, this theory is no longer tenable, "and it will be interesting to observe whether the incidence of trichobezoars will be lessened during this generation."

Various reasons have been given by some of these patients to explain their abnormal appetite. Schönborn's patient confessed after her operation that she swallowed her hair "to make her voice clear." Thornton's[3] patient had for years been in the habit of eating the combings of her hair and her sewing cotton to "clean her tongue." Jacobson's[7] patient swallowed hair because she "liked the tickling sensation" produced by the hair in its transit to the stomach. Paton's[8] patient ingested her hair "because it was so nice." Wren's[9] patient pulled out her hair and swallowed it in an unsuccessful attempt at suicide. According to Butterworth,[10] the habit of swallowing hair usually begins during childhood. It has also been observed that biting the finger nails is often a concomitant habit.

Hair balls are most frequently found in the stomach and only occasionally in the bowel. In only six of the 47 cases collected by Matas, that had been operated upon, were hair balls found in the intestinal tract. The tumor is formed by the matting together of the small bits of hair that have been swallowed, which, upon mixing with fatty material in the stomach, creates a solid mass. This mass often fills the gastric cavity and molds an exact cast of its interior. This cast when confined to the stomach describes the letter J; when prolonged into the duodenum it is S-shaped. In some cases other foreign bodies are found imbedded in the hair cast. As additional hair is swallowed, the mass may become very large. One reported by Davies,[16] the largest on record, weighed six and one-half pounds. As a rule only one hair ball is present, but occasionally two or more are found joined by faceted surfaces.

The clinical symptoms are largely dependent upon the size, position and the development of secondary complications. Frequently there will be no symptoms until the stomach is completely filled with the mass. Butterworth[10] states: "It is remarkable how little the general health suffers considering the duration of the habit. However, eventually the limit of toleration is reached and nature protests." The symptoms in general are those common to gastrointestinal upsets. Pain is a fairly constant symptom and usually nausea and vomiting occur at intervals. There may be alternating attacks of constipation and diarrhea. As the stomach fills with the mass the ingestion of food becomes more difficult, digestion more retarded and emaciation ensues. The breath becomes offensive and anemia grows more pronounced. Unless surgical measures are instituted, death will occur from inanition or perforation. Intestinal obstruction may also occur as a complication.[6, 38]

The preoperative diagnosis of trichobezoar before the advent of roentgenology was uncertain and seldom accurate. Of the 47 cases collected by

Matas, that had been operated upon, only ten had been diagnosed correctly prior to operation. Holland,[11] in 1913, published the first description of the roentgenographic appearance of a trichobezoar. O'Brien,[15] Rovsing[17] and others have made important contributions to the development of the roentgenologic diagnosis, and with modern technic, there should be no difficulty in making a correct diagnosis—in fact it is almost impossible to miss it. After administration of barium the mass can be seen lying free within the stomach, where it appears as a lighter area within the darker area of the barium which outlines and coats it. When viewed through the fluoroscope its position can be readily changed by palpation. A roentgenogram, five or six hours after the ingestion of the barium meal, is of the greatest value, the barium streaked hair ball showing plainly within the empty stomach. The history of swallowing hair over a long period of time, together with a palpable mass in the epigastrium, should certainly point to the possible presence of a trichobezoar. Frequently, however, the perversion is not admitted, but on the contrary is stoutly denied. The presence of bits of hair in the vomitus or stools is indisputable evidence.

The treatment of trichobezoar is exclusively surgical. There is no known medicinal agent that is powerful enough to dissolve the mass of hair that will not injure or destroy the gastric mucosa. The anterior gastrotomy incision, required in operating upon these cases, is often of unusual length, in order to release and remove the enormous mass without traumatizing the stomach wall. It is essential to exclude the intraperitoneal cavity if possible, and it is often difficult to deliver the greatly enlarged stomach and extraperitonealize it, so as to insure perfect asepsis. As Matas states, these hair balls are usually so foul and putrid, as the result of long-standing fermentation and decomposition of the hair masses which are mixed intimately with the residue of food of all kinds, that great care is required to prevent contamination of the peritoneum and abdominal walls with the drippings from the slick, slimy ooze that covers them. Drainage is not necessary unless contamination has occurred. Careful exploration of the intestinal tract is always indicated and enterotomy should be performed if hair balls are found in the bowel. In a case reported by Heazlitt,[13] intestinal obstruction occurred three days after a gastrotomy for the removal of a hair ball, and upon reopening the abdomen it was found that the obstruction was caused by a mass of hair in the ileum which had been overlooked at the first operation.

The results of operative treatment have been excellent. In 38 gastrotomies reported by Matas, not a single fatality occurred. In the remaining six of the operative cases in which hair balls were found in the intestinal tract and in which enterotomies were performed, there were two deaths. These operations were performed on nearly moribund children who had obstructions due to accumulation of hair in the ileum. Maes estimated the mortality to be not over 4 per cent. There was no operative mortality in the cases reported since 1928.

Case Report.—D. H., female, age 7, was admitted to the hospital, February 5, 1927, on account of a large tumor which was distinctly palpable in the epigastric region, accompanied by frequent attacks of pain, nausea and vomiting. Her mother stated that she had appeared to be a normal infant in all respects, but as soon as she started to crawl she began to pick up foreign bodies and swallow them. These consisted of hairs, strings, tacks, match sticks, pins and small pieces of glass, paper, chicken feathers and, in short, whatever she could put her hands on. Her mother found many of these objects in her

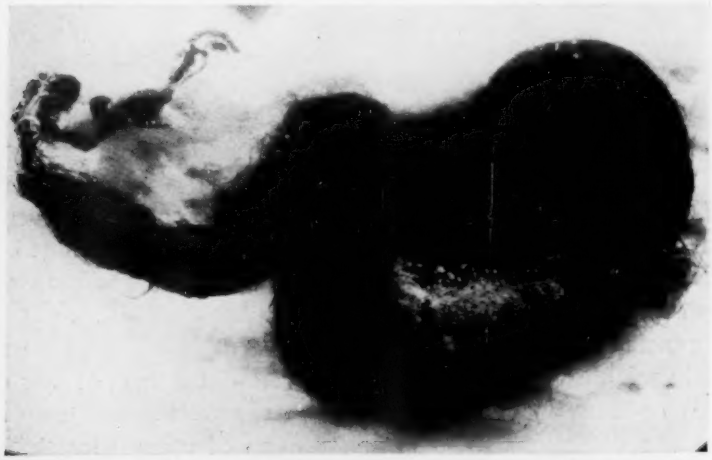

FIG. 1.—Photograph of hair ball removed, February 8, 1927, when patient was seven years old.

FIG. 2.—Roentgenogram of stomach containing the recurrent hair ball showing filling defect.

stools. At the age of two, she began to pull out her own hair and swallow it. This strange appetite continued, and, at the age of five, she began to lose weight and complained of pain in her upper abdomen. Her appetite for food was poor. These symptoms continued and gradually grew worse until nausea and vomiting supervened.

Physical Examination revealed a rather advanced state of emaciation. There was a large movable tumor in the epigastrium, which plainly outlined the contour of the

stomach. The front part of her head was devoid of all but very short stumps of hair. An attempt at roentgenologic examination was made and abandoned. She refused to swallow the barium meal, and was too irritable to cooperate in the taking of a simple exposure. However, in view of the history and physical findings the diagnosis was plainly trichobezoar.

Operation.—February 8, 1927: An anterior gastrotomy was performed and a large hair ball removed. This mass completely filled her stomach. There was a prolongation of it through the pylorus and duodenum into the jejunum for a distance of 13 inches. The pyloric opening was greatly dilated, but no gross pathologic lesions were encountered. This hair ball weighed one pound eight ounces and was S-shaped, forming a mold of her stomach, duodenum and part of the jejunum (Fig. 1). The mass consisted chiefly of hair, but many other foreign bodies were also evident.

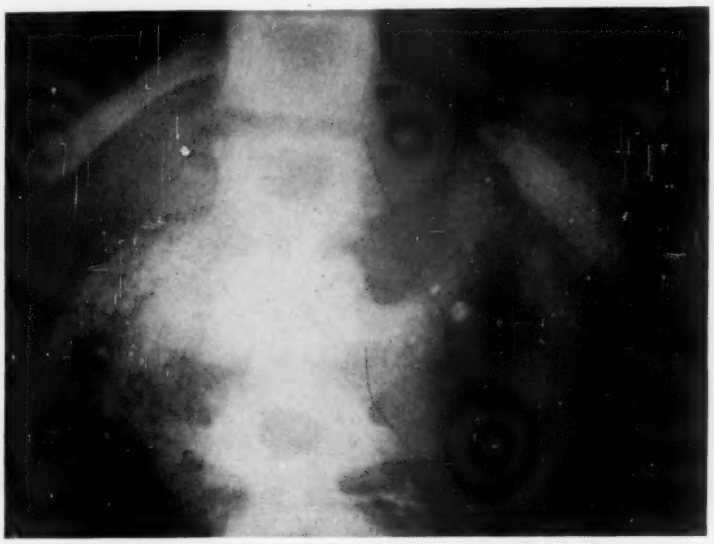

FIG. 3.—Six-hour roentgenogram showing barium streaked hair ball lying within the empty stomach.

Subsequent Course.—When she reacted from her anesthetic she began to beg for food. Her demands were so insistent and so noisy that it was necessary, for the comfort of other patients, to keep her completely narcotized until mouth feeding could be safely resumed. The patient made an uneventful recovery. She had a ravenous appetite and her digestion seemed to have been in no way impaired. During her two weeks' stay in the hospital she gained five pounds in weight and showed marked general improvement.

On January 18, 1935, eight years later, she was again admitted to the hospital on account of pain in her upper abdomen. She was now a girl, age 15, and had been attending high school until she was forced to leave school on account of her illness. Her teachers considered her unusually bright and intelligent. The pain in her abdomen was more or less constant but at times would become a severe colic. There was also a feeling of heaviness and fulness in her stomach accompanied by nausea. She was able to obtain some relief by inducing vomiting. These symptoms began about four months before admission. Prior to that time, she apparently had been free from symptoms since her operation in 1927. Her condition had grown steadily worse since onset of symptoms and her weight had dropped from 128 pounds in September, 1934, to 99 pounds upon admission.

Physical Examination revealed an emaciated, nervous, adolescent girl of normal mentality. The hair on her head was short in spots, especially near the forehead. There was a large epigastric mass that was freely movable and tender. W.B.C. 9,200, polys

70 per cent; R.B.C. 3,680,000, Hb. 72 per cent; Kahn test negative. She positively denied that she had swallowed hair recently, but after close questioning and being confronted with the nurse's statement that hairs were present in her stools, she admitted, reluctantly, that this perversion still existed. She also had the habit of biting her nails.

In the light of her history and physical examination it was plainly evident that she had accumulated another hair ball. Roentgenologic examination, after the administration of the barium meal, showed her stomach to be completely filled with a solid mass (Figs. 2 and 3). Her abdomen was again opened, January 20, 1935, and a large, twisted, S-shaped hair ball was removed. This hair ball weighed one pound two ounces and completely filled the stomach and duodenum (Fig. 4). It consisted almost entirely of black hair. In the first part of the jejunum, about seven inches from the duodeno-

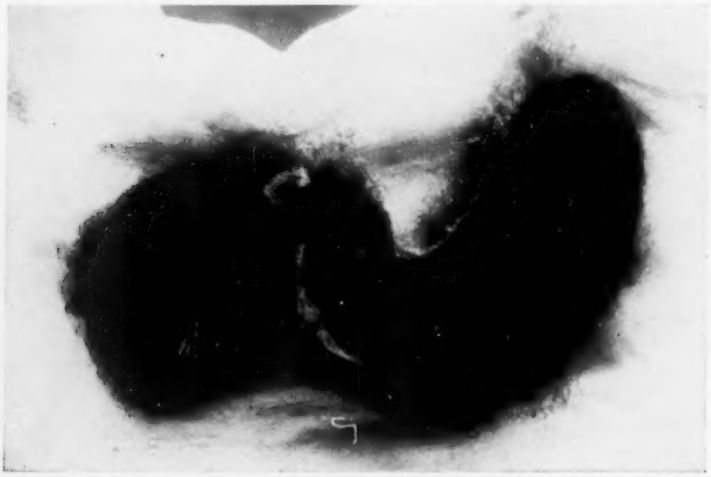

FIG. 4.—Photograph of recurrent hair ball removed, January 20, 1935, when patient was 15 years old.

jejunal junction, three small hair balls could be palpated. One of these had a very sharp edge which had ulcerated through the wall of the intestine. This perforation was covered by the omentum. A portion of the proximal end of the jejunum and its mesentery was much indurated and inflamed and there were numerous enlarged, inflamed mesenteric nodes in this region. With this extensive pathologic condition in the jejunum it was deemed advisable to resect the damaged portion rather than attempt to repair the perforation. Resection of the damaged segment and side-to-side anastomosis was, therefore, performed. The resected intestine contained the three small hair balls. Immediately distal to the inflamed portion of the jejunum there was an intussusception. This was, apparently, of recent origin and was easily reduced. The abdomen was closed without drainage and again the patient made an uneventful recovery.

SUMMARY

(1) Twenty-two additional cases of trichobezoar have been reported since the publication of Maes' paper, in 1928—making a total of 138.

(2) Recurrences are rare, as only three cases have been reported in the literature.

(3) A case with recurrence is reported herewith, in which perforation and intussusception occur as complications.

(4) The incidence of trichobezoars is practically confined to females, as only six cases have been reported in males.

(5) Roentgenologic examination is the most valuable agent for diagnosis.

(6) The treatment of trichobezoar is exclusively surgical.

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Discussion.—Dr. Ambrose H. Storck (New Orleans, La.): Doctors DeBakey and Oschsner have just completed a most comprehensive review of the world's literature concerning bezoars and concretions. The total number of cases which they were able to collect, including their own, was 311, 55.3 per cent of which were trichobezoars, 40.4 per cent of which were phytobezoars, and 4.1 of which were concretions. They have used the term diospyrobezoar to indicate specifically the persimmon bezoar, and this subclassification seems warranted because such bezoars constitute so large a proportion of all phytobezoars.

The incidence of trichobezoars was distinctly greater in the second and third decades, whereas, the incidence of phytobezoars was highest in the fifth and sixth decades. Over 80 per cent of the trichobezoar cases occurred in individuals under 30 years of age, but over 70 per cent of the phytobezoars occurred in individuals over 30 years of age. Individuals as young as one year old and as old as 56 years of age were found to have had trichobezoars, and the age range in the phytobezoar cases was from three years to 75 years. There is a striking difference in the incidence of trichobezoars compared to phytobezoars in respect to sex—91.4 per cent of trichobezoars occurring in females, whereas, females constituted only 23 per cent of the phytobezoar cases.

The clinical manifestations are likely to be vague, even though large and multiple bezoars are present. The discrepancy between the size of the bezoar and the degree of clinical manifestations is generally most striking in the presence of trichobezoars. Distinct or alarming symptoms do not usually occur unless complications develop and punctuate the indefinite symptomatology. Nausea and vomiting may result from obstruction produced by a bezoar and hematemesis may occur when a bezoar has caused the formation of an ulcer. Careful history taking in respect to the habit of hair eating, the eating of persimmons, or the ingestion of other substances likely to form a mass, may lead to a preliminary suspicion of the presence of a bezoar, but such

detailed history taking is usually not done. Likewise a history of the patient having passed hair in the stools would suggest the possibility of a trichobezoar's being responsible for otherwise unexplainable symptoms. It has been suggested that accurate diagnosis in the instance of gastric bezoars might be accomplished by means of direct examination by means of the gastroscope. Except for a sometimes detectable abdominal mass, the physical signs in the presence of bezoars are usually remarkably absent or indifferent. After all, in most instances the gastric bezoars depend upon the rather characteristic roentgenologic findings, obtained either by means of fluoroscopy or roentgenography. Even without the aid of contrast medium, it may be possible to detect an intragastric mass either by means of fluoroscopy or roentgenography. When a barium meal is administered, the spreading out of the radiopaque medium about the intragastric mass furnishes additional or further evidence of the existence of an intragastric foreign body. Subsequent roentgenologic examination, in the instance of trichobezoars, shows some of the barium remaining on the surface or in the interstices of the bezoar. Remaining flecks of barium may even indicate the extension of the bezoar into the duodenum. Insufflation of the stomach with air may be made, to more definitely demonstrate the lack of a sessile attachment of the tumor to the stomach wall. Even multiple intragastric masses may be detected or demonstrated roentgenographically and recognized preoperatively.

Aside from ulceration with the resulting hemorrhage at the resting site of the bezoar in the stomach, pyloric obstruction, perforation and intestinal obstruction are likely to occur as complications of bezoars. In some instances the primary manifestation of the existence of a bezoar has been the occurrence of intestinal obstruction resulting from a portion of an intragastric bezoar becoming detached from the main mass, while in other instances the obstruction has been due to an independent intestinal bezoar existing alone or in conjunction with a gastric bezoar. Because of their hard consistency, phytobezoars are more frequently associated with ulceration, hemorrhage and perforation than are the relatively soft trichobezoars. The finding of intestinal bezoars should be followed by an immediate or subsequent search for other intestinal or gastric bezoars.

The obvious advisability of applying surgical methods for relief of bezoars and complications occurring as a result of the presence of bezoars, is emphasized by a comparison of the mortality rates in the nonoperative and the operative cases. In the nonoperative group of trichobezoars, diosporobezoars and other phytobezoars, the mortality was 72.7 per cent, 50 per cent and 66.6 per cent, respectively; whereas, in the group subjected to operation, the respective mortality rates were 4.8 per cent, 7.4 per cent, and 4.7 per cent.

Dr. Frank S. Johns (Richmond, Va.): We have had two such cases which have been reported elsewhere. The first case, a woman, age 26, had absolutely no signs or symptoms of trichobezoars, except a mass in the upper abdomen. This patient's hair ball was a perfect cast of the stomach, and was easily removed, with no recurrence. The second case was that of a child, age 13, who came to the hospital with symptoms of obstruction, and a mass in the right side just above the head of the cecum. Her mother would not consider an operation, and took her home. She returned later with complete obstruction. She was explored promptly. A large abscess was found resulting from a large ball of hair in the terminal ileum and head of the cecum. The patient died shortly after operation.

Doctor Collins' case was particularly interesting, because of his patient's obviously high I. Q. combined with her habit of eating hair.

SURGERY IN HEMOPHILIA

REPORT OF FOUR CASES

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IN 1803, John Otto[1] published "An Account of an Hemorrhagic Disposition Existing in Certain Families," and, thereby, introduced hemophilia to the medical profession. We have, indeed, much earlier references to constitutional hemorrhagic tendencies which we would be justified in assuming to have been hemophilia. The most noteworthy is an account given by an Arabian author,[2] concerning a village of which the male inhabitants were liable to fatal hemorrhages from skin abrasions and other slight traumata. This report dates from the eleventh or twelfth century. Otto was, however, the first to call attention to what we now know as Nasse's law, that the disease manifests itself only in males and is transmitted by the females of the affected family as a sex-linked character. Nasse's[3] promulgation of this law did not appear until 17 years after publication of Otto's observations on the subject. What this law means is that the children of a sufferer from hemophilia will not show the disease, but his daughters and the female descendants of his children of either sex may transmit the disease to their sons. Apparently sporadic cases of hemophilia have been reported, and it is, of course, unsafe to rule out the condition because of the absence of a family history of the disease.

The exact nature of the hemophilic diathesis is still uncertain. The sole primary clinical symptom is hemorrhage. The major, if not the sole, demonstrated pathologic phenomenon is an abnormally slow coagulation of the blood. The bleeding time is generally normal. The blood cells are normal. The blood platelets are normal in number and size. The platelets show delayed clumping and disintegration; authorities are not agreed as to whether this is or is not important for the clotting time abnormality. Prothrombin and antiprothrombin, thrombin and antithrombin, blood fibrinogen and calcium in hemophilic blood have been assiduously investigated, and reinvestigated, sometimes with conflicting results. There is wide agreement that the blame for the protracted clotting does not lie in the blood fibrinogen or calcium, or in the anticoagulants present in the blood. Quantitative deficiency of prothrombin in the blood is the responsible factor in the view of Howell.[4] Addis'[5] experiments showed thrombin production to be normal in amount but delayed in time, and Mills[6] came to a similar conclusion, *i.e.*, "that the fault lay not in the amount of prothrombin or thrombin, but in the delayed activation of the latter by the former." He found the prothrombin in hemophilic blood much more resistant to activation with cephalin than that in normal blood. On the other hand, prothrombin once formed in hemophilic blood is the same both in amount and in action as that found in normal blood. Lenggenhager[7]

describes an experiment *in vitro*, which demonstrates the delayed formation of thrombin in hemophilic blood, the delay varying directly with the severity of the hemophilia; and he also found the final amount of thrombin formed to be the same in hemophilic as in normal blood.

According to Lenggenhager, this is not the solution of the hemorrhagic anomaly in hemophilia. He calls attention to the normal bleeding time, *i.e.*, the fact that the bleeding from small wounds, as from pricking of the ear lobe or finger tip, ceases as promptly in the hemophiliac as in the normal person, whereas, the bleeding from larger wounds is frequently so prolonged in the hemophiliac as to cause exsanguination. He is unwilling to accept the usual explanation, that small wounds close through the elasticity of the tissues, without the help of a blood clot. He states that the last blood to flow from a small wound always clots with great rapidity because thrombin is already present in large amounts, evidence that the bleeding is in process of being stopped by blood coagulation. Now this is the same in the hemophiliac as in the normal person. But if, after the spontaneous cessation of the bleeding, the small wound is pressed open, the bleeding which now begins does not cease promptly, but, under pressure, may continue for hours, in the case of the hemophiliac, whereas, it cannot be made to continue for longer than five to ten minutes in the normal person. In connection with this clinical experiment, Lenggenhager made the observation that, in the case of the normal person, clotting time decreased progressively with the duration of the bleeding, whereas, in the hemophiliac the contrary held: The first drops of blood to escape clotted in normal time, the later drops progressively slower; at the end of 45 minutes the blood which continued to flow from the ear lobe under the influence of pressure clotted almost as slowly as hemophilic blood from a vein. His explanation is that the thrombin first formed acts as catalyzer for the more rapid formation of more thrombin in the case of normal blood, but that this catalytic action is lacking or deficient in the case of the hemophiliac. Lenggenhager suggests that the use of the spring-lancet phlebotome, in obtaining blood for testing for coagulation time, may have been the cause of the paradoxical findings reported in the literature of almost normal coagulation time in hemophiliacs who, nevertheless, bled to death when operated upon.

I have taken for the title of my paper, "Surgery in Hemophilia," because I have four cases of serious surgical illnesses in hemophilic patients to report. I have observed a number of other hemophiliacs who had various hemorrhages into joints and muscles and from gums, kidneys and rectum. Operations should, it is admitted by all, be avoided in hemophiliacs. The lesser indications for surgical intervention are suspended and even in what we usually consider surgical emergencies, every effort is made to carry the patient safely through without resort to operation. Nevertheless, operation must sometimes be chosen as the lesser of two risks to life. In two of the four cases which I am reporting, operation was performed, and in two it was denied. In each of these groups, one patient survived and one succumbed.

CASE REPORTS

Case 1.—In August, 1921, W. R., age 19, consulted me for a tumor of the right breast, of six to eight months' standing. As given by the patient, the family history was negative; especially he gave no history of any member of the family having been a "bleeder." But the mentality of the family seemed low and I was unable to get any clear history from them after the operation. I made a diagnosis of carcinoma of the breast and proceeded to perform a radical breast amputation, removing both pectoral muscles and dissecting the axilla and closing the wound with a drain inserted through a stab wound in the side. During the operation I noticed that the bleeding was greater than usual.

About six hours after the operation, the skin flaps were markedly distended and a dirty looking, bloody fluid was oozing from the wound. I opened the wound a little so that the blood could be evacuated and gave the patient large doses of calcium lactate and several doses of horse serum. Before this medication his bleeding time was ten minutes and clotting time 16 minutes. The bleeding finally ceased and the wound healed rather slowly. The pathologic diagnosis of the specimen showed adenocarcinoma, without any metastases in the axilla.

Later this patient admitted that he had had hemorrhages before and had always had considerable trouble in stopping the bleeding whenever he cut himself or received any injury.

He continued to take calcium lactate for some time after leaving the hospital. I saw him at intervals for 10 years following the operation, and have seen him in the last month. He had been entirely normal and had had no trouble except small hemorrhages into several of the joints.

Case 2.—In January, 1931, Robert S., age 25, was admitted to the hospital, with indications of an abdominal emergency. His mother died in his childhood and an adequate family history was not obtained. He stated that he had been found to be a bleeder when he was two years old and that he had been sick all his life. He had had hemorrhages from the bladder and kidneys and bleedings into several joints. Six years before he had an attack of appendicitis, from which he recovered without operation. On the day before admission, he had an attack of abdominal pain, followed by nausea and vomiting. On examination, the abdomen was markedly tender and rigid. The urine contained red blood cells. The leukocyte count was 18,600 with 90 per cent polymorphonuclears. The bleeding time was three minutes, the coagulation time six minutes. He appeared acutely ill, and I believed there was perforation of the appendix with spreading peritonitis.

Appendicectomy was performed under a general anesthetic. A gangrenous, perforated appendix was found, with free pus in the peritoneal cavity. Particular care was taken to clamp and ligate all vessels and the patient was given 500 cc. of citrated blood by transfusion after the operation. His coagulation time was the same as before the operation.

For five days his condition remained very good. He was given quantities of dextrose solution intravenously, and a second transfusion of 500 cc. of citrated blood. He also received fibrinogen. There was no bleeding from the wound, nor was the wound swollen. On the sixth day, the patient did not look so well. He complained of abdominal pain and had considerable bleeding from the wound. The wound edges were distended. The coagulation time was now increased to ten minutes. He was nauseated and vomited. He was given a third 500 cc. of citrated blood and more dextrose solution intravenously. The wound became distended with blood clots and opened up throughout. The whole abdomen was markedly distended and rigid. The patient died on the ninth day after the operation.

The patients in Cases 3 and 4 were brothers. It was stated that the maternal grandfather may have been a hemophiliac, but the mother was not certain of this. I did not see the patient in Case 4 during his fatal illness, as he was at that time living in another city. But his physicians consulted with me and were kind enough to supply me with the data.

Case 3.—In October, 1935, M. H., age 19, was admitted to the hospital because of

abdominal pain. Since the age of eight months, he had been known to have hemophilia. He had had numerous hemorrhages into the joints and into the tissues of the neck, and at one time hemorrhage had followed suppuration of the sublingual glands. For the past few years, he had been under the care of Dr. C. A. Mills, of Cincinnati, and I had seen him many times with hemorrhages and injuries. On the night of October 26, 1935, he was seized with sudden severe abdominal pain, which localized in the right lower quadrant. He was nauseated, but did not vomit. There was much tenderness in the appendiceal region and rigidity of the right abdomen. No mass was made out. The leukocyte count was 22,300, with 90 per cent polymorphonuclears, 17 per cent nonsegmented cells. Coagulation time, Biffi Brooks, ten minutes; test tube, 18 minutes.

It was decided that the patient had a better chance for recovery without operation, and Doctor Mills' advice was sought and treatment as recommended by him was carried out. The patient was given 1 cc. theelin and fibrinogen each day, nothing by mouth, and fluids parenterally. A blood count was made every day. On November 1, 1935, the blood count was down considerably and he was given liquids by mouth. A definite mass could be palpated in the right lower abdomen, which seemed to me to be a walled-off abscess.

November 8, 1935, about 11 days after the beginning of his attack, the patient had acute abdominal pain and a short time afterwards evacuated large tarry stools. His temperature and leukocyte count came down. During the following two weeks, he continued to pass large amounts of tarry stools, and his red cells went down to 1,610,000, hemoglobin 24.2 per cent, and there was considerable reduction of the leukocyte count. Of course, he received frequent and large blood transfusions during this time.

At the end of two weeks, he improved greatly and the bleeding from his intestine ceased. He was now able to take nourishment normally and, with the aid of more blood transfusions, his hemoglobin and red blood cells had returned to normal by December 8, 1935, and his leukocyte and polymorphonuclear count was also normal. He was eating well and his bowels moved well. His abdomen was soft except in the right lower quadrant where he had a large and tender mass.

I have observed him frequently since his discharge from the hospital. His bleeding time has ranged from two to ten minutes and his clotting time from six to eight minutes. The mass in the right lower quadrant persisted for nearly a year. Now it cannot be palpated at all. Since his attack of appendicitis, he has had hemorrhages into several joints and bleeding from the kidneys; however, he has carried on his school and other work, though with some difficulty, and this year graduated from the University. At times he has been given courses of theelin and fibrinogen and kept on a high protein diet.

Case 4.—H. T. H., Jr., age 25, older brother of M. H. (Case 3), had been known all his life to be a bleeder, but had had no serious hemorrhages except small ones into several joints and from the gums.

After his graduation from the University he moved to an Eastern city. He seemed to be in perfect health when he left Lexington, but a few days after his arrival there he had an acute attack of appendicitis. He understood all about himself, as he had seen his brother go through the attack which I have described. He was taken to an excellent hospital and cared for by excellent surgeons and medical men. He was treated by blood transfusions, theelin and fibrinogen, with fluids parenterally and nothing by mouth. He was not operated upon. His appendix ruptured and the infection did not wall-off. He died of generalized peritonitis at the end of eight days.

The situation of the hemophiliac has changed greatly for the better in the last few years, and this is particularly true of the hemophilic individual who must undergo a surgical operation. To-day it is possible to prevent and to control hemophilic hemorrhages, whether spontaneous, accidental or post-operative, to a degree that was wholly impossible previously.

Of the 1,000 cases (approximate) which Carrière[8] collected from the medical literature, in 1907, 89 per cent had terminated in fatal hemorrhage

before the subjects had reached the age of 21. But, in 1929, Weil[9] felt able to assert that one no longer had the right to let hemophiliacs die of hemorrhage. The most dangerous period in the life of the hemophiliac lies between the age of one year and adolescence. If he can be brought safely to manhood, there is a chance that the hemorrhagic tendency may lessen to a considerable degree.

It is important to note that the clotting time of the blood of the hemophiliac is liable to wide variations for no known reason. These swings are equally sudden in both directions, and it would, therefore, be highly unwise to assume safety in operating upon a known or suspected hemophiliac because his clotting time at some recent period had been relatively close to normal. While on the subject of cautions, a word might be said as to the danger of mistaking the symptoms of hemophiliac hemarthrosis for rheumatism and proceeding, thereupon, to tonsillectomy or tooth extraction. Another caution worth repeating is not to let slow bleeding or oozing of blood go on for a day or two with the expectation that, being slight, it must soon stop of itself. Conrad[10] tells of a hemophilic child who was allowed to ooze blood for six days after an operation before any energetic measures were taken. The child was then too far exsanguinated to respond to blood transfusions.

There is no known curative treatment for the hemophilic diathesis, but it has proved possible, in a few reported cases, to bring about a certain inactivity and maintain this improvement by repeated treatments. Weil[9] has been able to follow, over a period of years, 20 patients treated monthly or every two months by subcutaneous injections of serum (probably horse serum), none of whom has had serious hemorrhages during this time. The injections reduce coagulation time. One of his results was particularly brilliant: A member of a notable hemophilic family, which has produced more than 200 cases of hemophilia since the beginning of the eighteenth century (the Tenna family) was given preventive serum injections regularly for seven years; ten years after the last injection he had had no return of hemorrhages and his blood coagulation time was still normal.

Ovarian extract therapy has also been used in the hope of benefiting the basic condition. Birch[11] reports favorable results in 19 cases followed for six months. The treatment must be continued indefinitely. Unfortunately, a number of other investigators report failure with the method. Ovarian implants have been used with partial success in a very few cases. Birch used a preparation of the follicular hormone, theelin, injected subcutaneously every second or third day. In two or three weeks the clotting time is reduced nearly to normal, where it can be kept by an injection twice weekly. Mills[6] points out that a field of usefulness for this form of treatment might be in the preparation of a patient for tooth extraction or minor surgical procedures.

Other measures have been recommended, particularly for the preparation of a hemophilic patient for a necessary operation, the object being to shorten the clotting time temporarily. Mills gives first place in prophylaxis to sensitization to a foreign protein. He states: "It is effective in such a large percentage of hemophiliacs that no physician handling these patients is justified

in failure to make proper use of it." The idea is to sensitize the patient by intramuscular injection of a foreign protein, preferably sheep or hen serum, after which intradermal injection of a drop of the serum to produce local reaction (the usual urticarial wheal) will be followed by a marked improvement in the coagulability of the blood. The production of a generalized systemic protein reaction must be avoided, since this might increase the hemorrhagic tendency disastrously. Mills states: "By keeping all one's hemophilic patients sensitive to some foreign protein . . . one always has at hand an immediately effective method of treating hemorrhages as they occur. And if one desires to use the method for the prevention of bleeding, one needs only to induce the skin reactions as often as is indicated by observations of blood coagulability."

Mills also recommends a high protein diet in prophylaxis and in treatment of the hemorrhages. By a regimen containing some protein at each meal and milk, eggnog, *etc.*, between meals and once during the night, the protein effect on the blood coagulability, he states, is continuous and tends to be cumulative.

Vitamins, especially B, C and D, have been used with success in some cases and failure in others, in the effort to shorten the clotting time. Havet[12] states that the mechanism of vitamin effect in hemophilia is uncertain. There may be an action on the vessel walls; one of the theories of the etiology of hemophilia includes abnormal fragility of the vessel walls. Or it may be that an improvement is effected in the metabolism of calcium in the case of vitamin D. Vitamin C is given intravenously. The vitamin complex Nateina Llopis is given by mouth. Blood transfusion is generally recognized as the surest method of preventing, as well as of checking, hemorrhage in hemophilia. Transfusion fulfills a double function: it furnishes the principal coagulants in which the blood of the hemophiliac is deficient, and it supplies blood volume to replace blood loss. Both pure blood and citrated blood are used.

The reduction of the clotting time brought about by transfusion is of brief duration—about 24 hours, according to Feissly,[13] who was the first to publish success with the method—but the effect, when it wears off, may be renewed by a repetition of the transfusion. A transfusion may be given the evening before operation. Bertrand-Fontaine and l'Hirondel[14] feel that transfusions of 200 to 300 cc., carried out one hour before surgical intervention, will almost surely prevent postoperative hemorrhage if hemostasis is possible, especially if the patient has been given from 20 to 40 cc. of serum intramuscularly on each of the two days preceding the operation.

As to the technic of operation best suited to interventions on hemophiliacs, Firor and Woodhall[15] used an electrosurgical unit for the amputation of the thumb in a case of acute hemarthrosis simulating "bone sarcoma" in the roentgenogram. Blood loss was minimal and the healing process was essentially normal. These authors feel that the value of the electrosurgical technic "can scarcely be over estimated both for major surgical procedures on these (hemophilic) patients and for the control of hemorrhage from small lacera-

tions." Blalock[16] employed piecemeal tissue ligation with catgut ties in an amputation of the arm in a hemophiliac, the ligating process including "every bit of soft tissue except the skin." A slow but steady loss of blood continued for more than two weeks following the operation, in spite of direct transfusions given approximately every third day.

The attack on the hemorrhage itself should be by both general and local measures if the site of the bleeding can be reached. The usual measures of stopping bleeding, such as ligation of bleeders and compression, will, of course, be used, but measures directed to increasing the coagulability of the blood should also be applied at once, and in conjunction, not tried out one at a time. It cannot be too strongly emphasized that a "wait and see" policy has no place in treating hemophilic bleeding.

Unless the patient was under hospital care at the time the hemorrhage started, as in the case of bleeding following operation, much time is likely to have elapsed before the physician has the opportunity to start treatment. With great truth, Mills says: "The first two days offer the greatest chance of prompt stoppage of the bleeding. Beyond this period great difficulty may be encountered and repeated blood transfusions required."

For reasons of cost and because of the fact that the patient sometimes takes the preparation for blood transfusion as a signal for alarm, Mills does not advise transfusion during the first two days of bleeding, when other forms of treatment are usually effective. But by many, transfusion is regarded as the first line of defense in any serious hemophilic bleeding. It is recognized by all as the method of choice in prolonged hemorrhages. Transfusion usually checks the bleeding rapidly, but may have to be repeated. Intramuscular serotherapy, 20 cc. of some human or animal serum, such as antidiphtheritic serum, as fresh as possible, may be expected to act in from 12 to 24 hours. Kimm and Van Allen[17] recommend intramuscular injection of extract of the whole ovary, 4 gr. every six hours until the hemorrhage is controlled. Ovarian therapy in hemophilia is based on the assumption, not yet wholly confirmed, that the female hormone is totally lacking in the blood of hemophilic males.

Mills favors, among biologic coagulants, purified tissue fibrinogen, administered orally in 3 to 5 cc. doses (of a 1.5 per cent solution) in cold water, on an empty stomach, preferably one-half hour before meals and at midnight, or injected subcutaneously in doses of 1 to 2 cc. every two hours, changing to every eight hours after four doses. In treating hemorrhage by any biologic agent, it is important to continue the treatment for at least 24 hours after bleeding has ceased. Mills offers a special caution as regards the use of transfusion in connection with tissue fibrinogen treatment: Transfusion must not be carried out within eight hours of a previous dose of fibrinogen. However, it may be administered one hour after a transfusion. In the case of a patient previously sensitiz 1 to a foreign protein, one has at hand an excellent means of checking hemorrhage in the induction of the skin reaction, as already mentioned. Adrenalin by subcutaneous injection is useful if the hemorrhage is not arterial. Witte's peptone given subcutaneously, extract of platelets, calcium chloride given intravenously, and sodium citrate administered by the

same route, all find sponsors in the literature. Injection of blood into the buttocks has been followed by good results. Lucas[18] injected 10 cc. of the mother's blood subcutaneously in a newborn infant seen with a severe hemorrhage from the intestines. The bleeding diminished markedly and was later controlled by hemoplastic serum.

Eley[19] calls attention to the hemostatic value of human placental extract. He states that experiments *in vitro* suggest some degree of specificity for human as against bovine tissue extracts. He treated 20 hemophilic patients with human placental extract at the Children's and Infants' Hospital, Boston. In 13 cases, the coagulating time of the venous blood was reduced to ten minutes, and some reduction was obtained in three other cases. In four cases, the treatment failed to influence the clotting time. He found the oral preferable to the intramuscular route, since the extract was effective in from 20 to 30 minutes when given orally and required several hours to show an effect when injected into the muscle.

For topical application, Eley[19] used a sponge soaked in sterile placental extract applied over the lesion after the edges of the wound had been brought together with adhesive tape. Bleeding ceased and a firm clot developed.

Lenggenhager reports constant success from bringing into the wound a concentrated thrombokinin (or thrombokinase), prepared by boiling for three minutes finely chopped fresh human or animal struma in three to five parts of physiologic solution, and filtering. A pledget is dipped in this solution and applied firmly to the wound for several minutes or the wound is tapped lightly with the saturated pledget several times, so that the solution is pressed into all parts of the wound. He recommends injecting this solution into the operative field along with the anesthetic solution previous to operation. To stop bleeding from the gums he injects the thrombokinin solution together with adrenalin into the bleeding area. He has seen this solution stop bleeding after tooth extraction when all other methods failed.

A compressive dressing moistened with human or animal serum and applied to the wound after proper cleansing is often effective in the prompt control of hemophilic hemorrhage.

The treatment of an infected wound in a hemophilic patient is a tedious process, the continual danger of hemorrhage making great caution necessary. The successful use of maggots in the treatment of a large infected wound, in a case of severe hemophilia, was recently reported by Pohle and Maddock.[20] Their experience seems to indicate that when active bleeding has ceased, maggots can be applied without danger of exciting fresh hemorrhage.

SUMMARY AND CONCLUSION

Four cases of serious surgical illnesses occurring in hemophilic individuals are reported. Two were treated by operation, with one recovery and one death. Two were treated without operation, likewise with one recovery and one death.

The hemophilic individual should not be subjected to operation unless refusal of operation would place him in still greater danger to life.

Safeguards should be thrown about the hemophilic patient who must

undergo a surgical operation, by having recourse to the modern biologic methods of decreasing temporarily the clotting time of the hemophilic blood, such as blood transfusion, serum therapy, ovarian therapy, intradermal injection of a drop of serum to which the patient has been sensitized, administration of tissue fibrinogen, *etc.*

While these methods may not succeed in every case, nevertheless, applied in time and in proper combination, they have frequently proved life-saving.

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